



THE UNIVERSITY *of* EDINBURGH

This thesis has been submitted in fulfilment of the requirements for a postgraduate degree (e.g. PhD, MPhil, DClinPsychol) at the University of Edinburgh. Please note the following terms and conditions of use:

This work is protected by copyright and other intellectual property rights, which are retained by the thesis author, unless otherwise stated.

A copy can be downloaded for personal non-commercial research or study, without prior permission or charge.

This thesis cannot be reproduced or quoted extensively from without first obtaining permission in writing from the author.

The content must not be changed in any way or sold commercially in any format or medium without the formal permission of the author.

When referring to this work, full bibliographic details including the author, title, awarding institution and date of the thesis must be given.



THE UNIVERSITY *of* EDINBURGH

**A brief group intervention to support people with Functional
Neurological Disorder in the Acute Sector: A feasibility study**

Pauline M Insch

Doctorate in Clinical Psychology

University of Edinburgh

July 2020

Acknowledgements

First of all, I would like to thank my clinical supervisor, Dr Fiona Summers, for introducing me to Functional Neurological Disorder. I'd also like to thank Fiona for her guidance, patience and support which have been invaluable throughout this whole process. I have been inspired by her drive and passion as a clinician and will remember my time working with her fondly. I would also like to express my gratitude for my second clinical supervisor, Dr Ceri Trevethan, for her help with the planning of the project in the early stages. Thanks are also extended to Dr Clara Calia, my academic supervisor, for her enthusiasm and encouragement in addition to her prompt responses and feedback on draft chapters. Special thanks to all the participants who gave up their time to find out about and take part in the research. Their efforts were much appreciated as was their feedback and commitment to making a difference, not only for others living with FND but also the clinicians supporting them.

Thank you to everyone in the Department of Clinical Neuropsychology for their interest in and support of the project and for making me feel so welcome. In particular Mrs Joaane Coull, for updating the participants visit schedule every week and Mr James Anderson for his advice and sharing his office space every Friday. Also thank you to Beccy Harragan, for her enthusiasm and for everything she did to help me organise the groups. Special thanks to Dr Adele Simpson for her encouragement and kindness towards the end of the process. I would also like to acknowledge the support I have had from the Department of Neurology in recruiting for this project. I am particularly grateful to Dr Elizabeth Visser and Dr Callum Duncan for taking time to help me put together the information videos used in the groups.

I am also grateful for the support received from Professor John Crawford, Professor Louise Philips and Dr Mary Kilonzo from the University of Aberdeen. Thank you to Dr Gillian Slessor for proofreading my draft and her continuing friendship. I would also like to thank Dr Liam Mooney for his support and friendship but also for being my second rater on the systematic review chapter, his efforts were much appreciated especially as he was completing his own thesis at the time.

I would like to say a personal thank you to friends, old and new, for seeing me through, not just this thesis but the experience of clinical training, your faith in me has been much appreciated. Last, but by no means least, thank you to my husband and son, Stuart and Lewis. Your love, support and faith in me has kept me going throughout this training journey. Finishing my training in the height of a global pandemic has not been ideal, but I can't think of no one else I'd rather face lock down with and look forward to our next chapter of adventures together.

Table of Contents

Acknowledgements	2
Table of Contents	3
Portfolio Thesis Abstract.....	5

Chapter One

Systematic Review Abstract.....	7
Introduction.....	8
The Self-Regulation Model (SRM).....	12
Outcome Measures Based on the SRM.....	16
Health Beliefs in FND.....	17
Review Aims and Objective	18
Method	19
Inclusion and Exclusion Criteria.....	19
Screening	21
Eligibility	21
Included	21
Identification	21
Literature Search Strategy	22
Quality Assessment.....	23
Results	26
Study Characteristics	26
Applying the Newcastle Ottawa Scale Quality Criteria	32
Narrative Synthesis	37
Discussion.....	41
References.....	45
Chapter 1: Appendices	53
Appendix 1 Submission guidelines for Neuropsychology review.....	54
Appendix 2: Adapted Newcastle-Ottawa Scale	59

Chapter Two

Empirical Paper Abstract	62
Introduction	62
Method.....	62
Results.....	62
Discussion	62
Introduction.....	63
Summary	71
Study Aims	73
Methods.....	74
Participants	74
Design.....	75
Recruitment Procedure.....	76
The Group Intervention.....	77
Group Resource Development.....	78
Group Outcome Measures	80
Ethical Approval.....	89
Statistical Analysis.....	89
Results	92
Study Aim 1.....	94
Study Aim 2.....	108
Study Aim 3.....	111
Study Aim 4.....	117
Discussion.....	123
Strengths and Limitations.....	131
Future research.....	132
Conclusions	134
References	135
Chapter 2: Appendices	144

Portfolio Thesis Abstract

Purpose

Chapter one used the Self-Regulation Model (SRM) of health beliefs as a framework to review results from psychological interventions that utilised health belief outcomes based on the SRM for people with Functional Neurological Disorder. The empirical chapter used demographic, psychological and healthcare utilisation outcomes to explore the efficacy, feasibility and acceptability of a brief intervention for routine care for people with FND designed to address a gap in current service provision within NHS Grampian.

Method

A systematic search strategy identified seven studies for inclusion in the review chapter.

Chapter two compared the data of attenders (n=16) and non-attenders (n=13) on demographic, psychological and healthcare utilisation variables. Data from group completers was collected at four timepoints and included cognitive function, mood, quality of life, health beliefs, healthcare utilisation and costs; Participant knowledge and experiential data was also collected.

Results

Chapter one showed studies were of mixed quality with studies being rated as satisfactory (n=4) unsatisfactory quality (n=3). Higher quality studies had more comprehensive selection and data management processes than lower quality studies. In chapter 2 non-attenders rated their physical health limitations as significantly more and perceived less personal control than attenders. Completers showed significant improvements in understanding of FND and in levels of fatigue were reported as significantly less at follow up. No differences between attenders and non-attenders for healthcare utilisation or costs. 63% of completers showed improved health status at follow up, participant expectations were met. knowledge of FND significantly improved. The content and format of the intervention were acceptable to participants

Conclusions

Taken together both chapters highlighted the role of health beliefs in FND but also the importance of providing access to appropriate information and peer support for those receiving a diagnosis. The brief intervention showed promising results in terms of efficacy, feasibility and acceptability. Although further research will be required to ascertain if these findings are sustainable in a larger sample long term.

Word Count (excluding references and appendices): 27, 382

**Exploring the impact of psychological interventions on the health beliefs
of patients with Functional Neurological Disorder: A Systematic Review**

Pauline M Inch * ^{1,2} Clara Calia ² and Fiona Summers ¹

¹ Department of Clinical Neuropsychology, NHS Grampian. Aberdeen, Scotland

² School of Health and Social Science, The University of Edinburgh, Scotland

*Corresponding author

E-Mail: S17947031@sms.ed.ac.uk

Orcid ID 0000-0001-8824-906X

Written in the style for submission to *Neuropsychology Review*

Word Count (excluding references and appendices): 10, 328

Systematic Review Abstract

Introduction

This systematic review aimed to explore the pattern of health beliefs of people with Functional Neurological Disorder (FND) after taking part in psychological interventions as indexed by measures based on the self-regulation model of health beliefs.

Methods

A systematic review using a pre-determined search strategy was undertaken. Searches of electronic databases and sources of grey literature were searched from January 1995 to January 2020. In addition, references of included studies as well as studies citing the included studies were manually searched.

Results

Seven studies were included in the review, studies were evaluated against quality criteria based on the Newcastle Ottawa Scale. Results showed four studies met criteria for rating as satisfactory and three studies were found to be of unsatisfactory quality. Overall studies rated as higher quality were found to be most robust in their selection processes, utilising relevant statistical analysis and made efforts to identify and control for confounding variables. Studies rated as poorer quality tended to detail in justifying their methodology and lack specificity when reporting and analysing outcome data.

Conclusion

There was provisional evidence to suggest psychological interventions have potential to change the maladaptive beliefs those with FND may have about their symptoms. However, the methods around analysis and reporting of health belief outcomes in non-randomised research lacked consistency.

Keywords

Health beliefs; Illness perception; Functional Neurological Disorder; Psychological interventions; Self-regulation Model;

Introduction

Functional Neurological Disorder (FND) refers to the physical manifestation of neurological symptoms that have no organic basis as there is no detectable damage to the nervous system. Symptoms are real, not imagined and can be as debilitating both physically and psychologically as organic disease (Stone, Carson and Sharpe, 2005). The terminology used to describe FND has been a source of debate in the past with the condition being referred to as hysteria, dissociative, psychogenic, psychosomatic, somatoform, conversion and medically unexplained (Edwards, Stone & Lang, 2014). More recently there has been a shift in labelling these neurological symptoms as functional which is thought to remove speculation around the aetiology of presenting symptoms (Edwards et al., 2014). The use of the label 'functional' appears not only to be perceived as less offensive to those receiving a diagnosis but is also being adopted as the term of preference for clinicians treating those living with symptoms (Rommelfanger, Factor, LaRoche, Rosen, Young & Rapaport, 2017; Stone, Wojcik, Durrance, Carson, Lewis, et al., 2002).

There is a lack of epidemiological research around FND which may be due, to the complexity of categorising symptoms clinically and the diagnosis process (Carson & Lehn, 2016). As symptoms can be varied there has been debate historically regarding how to conceptualise and categorise the symptoms (Carson & Lehn, 2016). Although this appears to be improving with a focus, in the DSM V, on patterns of symptoms that cannot be attributed to organic neurological conditions (Carson & Lehn, 2016). As a result of this complexity, FND can be difficult to diagnose without extensive experience of neurological conditions (Carson & Lehn, 2016). For example a study investigating a diagnosis of FND made by General Practitioners (GP) found that the accuracy of diagnosis was at chance level (Carson, Ringbauer, Stone, McKenzie, Warlow & Sharpe, 2000) whereas those diagnosed by Neurologists were more robust over time (Stone, Carson, Duncan et al., 2010). This

discrepancy raises the importance of the level of clinician training and knowledge levels when accurately diagnosing FND (Carson & Lehn, 2016). As a result the most valid figures for prevalence of FND should be taken from settings where neurologists are undertaking the diagnosis which may make the cost of multi-centred, worldwide epidemiological studies prohibitive (Carson & Lehn, 2016).

There is a limited range of research available exploring the prevalence of FND within a Neurology clinic setting with the majority of research being based in the UK finding prevalence rates of FND between 26% and 45% (Bateman & Harrison, 2000; Carson et al., 2000; Stone et al., 2010; Stone & Sharpe, 2002; Stone, Sharpe, Deary, I., & Warlow., 2004). There is also some variability within international research studies. For example a study in the Netherlands found 35% of patients presented with medically unexplained symptoms where as an Australian study showed 15% of outpatients presented with FND (Ahmad & Ahmad, 2016).

In Scotland around one third of patients presenting to neurology clinics based have FND related symptoms (Carson et al., 2000; Stone, Carson, Duncan, Roberts, Coleman et al., 2012; Stone & Sharpe, 2002; Stone et al., 2005). FND can present in many forms including non-epileptic attack disorders, tension headache, limb weakness, abnormal movement, cognitive and sensory problems. It is common for patients with FND to have comorbid physical and mental health conditions and/or a functional overlay to symptoms of neurological conditions such as epilepsy (Stone et al., 2012).

Patients with FND are the largest clinical group accessing neurology clinics. In Scotland, 5,000 people per year receive a diagnosis of FND, this is important for healthcare services as the healthcare utilisation of patients with FND is estimated at double that for those without (Healthcare Improvement Scotland, 2012). Estimated costs for healthcare service providers within Scotland are £1.3 million for outpatient services, £6.01 million for

inpatients and £4.01 million for primary care services (Healthcare Improvement Scotland, 2012).

In addition to scheduled care, there is evidence people with FND present regularly for unscheduled care such as the emergency room (Cock & Edwards, 2018). Guidelines for treating and managing symptoms of FND have recently begun to emerge. Healthcare Improvement Scotland (2012) recommend a 4-level stepped care model starting with a robust explanation and validation of the FND diagnosis through to step 4 with patient pathways and clinician training. This report also highlighted a lack of consistency relating to evidence based therapeutic interventions in FND. The inconsistency came from the heterogeneity of delivery methods and patient outcome measures which made it difficult to generalise findings across studies (Division of Neuropsychology, 2013).

There is tentative evidence that health beliefs are an important predictor of progress 12 months later for those with FND (Sharpe, Stone, Hibberd, Warlow, Duncan et al., 2010). A potential mechanism for the development of beliefs around our subjective health comes from both personal experience and external information from the environment (Hotopf, Mayou, Wadsworth, & Wessely, 1999). In childhood the misinterpretation of symptoms which is reinforced by parents can lead an individual to believe that their perceived health is poorer than their actual health (Benjamin & Emminson, 1992). This pattern of thinking can continue into adulthood resulting in an inaccurate concept of current health state (Benjamin & Emminson, 1992). Environmental information can also be acquired in a secondary manner, for example those with FND often have experienced family health problems (Hotopf et al., 1999). Information from the media about symptoms relating to illness can also be internalised and result in higher numbers of people presenting with concerns they have the condition in question (Stewart, 1990). A potential mechanism for maladaptive beliefs may result from both personal experience and secondary sources of information contributing to

the beliefs an individual accesses when making sense of the symptoms they are experiencing (Hotopf et al., 1999).

There is also evidence that patients with FND hold a significantly stronger belief that psychological factors do not contribute to their symptoms when compared to patients with organic neurological conditions (Stone, Binzer & Sharpe, 2004). This belief that symptoms originate from an organic source has been identified as a potential defining feature of FND previously, however the dismissing of psychological factors may also be an artefact of the diagnostic process where a medical explanation for their symptoms that the patient finds credible has not been achieved (Stone et al., 2004). This is important as there is also evidence to suggest those with negative health beliefs are less likely to be reassured when results of medical tests come back normal (Donkin, Ellis, Powell, Broadbent, Gamble & Petrie, 2005) which may lead to increased healthcare utilisation searching for the 'right' diagnosis.

Psychologically based treatment programmes have been identified as being a potential mechanism in helping patients reframe their beliefs previously (Stone et al., 2004). However, there appears to be a disparity in the literature with emerging evidence from related clinical guidelines that psychological interventions are an appropriate treatment but people with FND have difficulty engaging in mental health related services (Monzoni, Duncan, Grunewald & Reuber, 2011). The language used in the explanation given as to why there is a lack of physical source for the symptoms has been perceived by those receiving the diagnosis as their symptoms being dismissed as imaginary or fictitious (Stone et al., 2002). This interpretation can mean patients resist acceptance of an FND diagnosis covertly by not engaging with services or more overtly, by disputing the diagnosis (Monzoni et al., 2011). This behaviour could potentially be understood in terms of the Self-Regulation Model (SRM) with the refusal to attend mental health services due to the beliefs a person with FND has constructed about their illness. Sharpe et al. (2010) utilised items from the Illness Perception

Questionnaire in their study (Weinman, Petrie, Moss-Morris, & Horne., 1996). This is an outcome measure derived from the SRM (Leventhal, Meyer & Nerenz., 1980; Leventhal, Nerenz, & Steele.,1984) that theorises that health beliefs are important factors contributing to the level of symptoms being experienced as part of an illness.

The Self-Regulation Model (SRM)

In the SRM the person experiencing the symptoms operates as an active agent. The processes within the SRM occur simultaneously rather than in stages (Leventhal et al., 1980; 1984). For example, when a person responds to a perceived threat to their health, a response to assist an understanding of the symptom becomes activate which allows meaning of the symptoms to be constructed. There are several factors thought to contribute to the patterns of meaning people perceive when facing a health threat according to the SRM (Leventhal et al., 1980; 1984).

The first component in the SRM is the cognitive representation that is developed by the individual. In the SRM, this cognitive representation underpins several other factors related to the illness experienced. Illnesses and disorders have a diagnostic label attached, some illnesses are culturally accepted and have no positive or negative connation's for most people e.g. the common cold or influenza. However, sometimes labels are stigmatised in society and if a label from an illness with negative connotations is attributed to a person, they may internalise these. This process of responding to the diagnostic label and the attached societal meaning given to a health condition is called illness identity in the SRM model (see Figure 1). According to the SRM, any response to the label assigned will be based on the personal understanding of what the condition is.

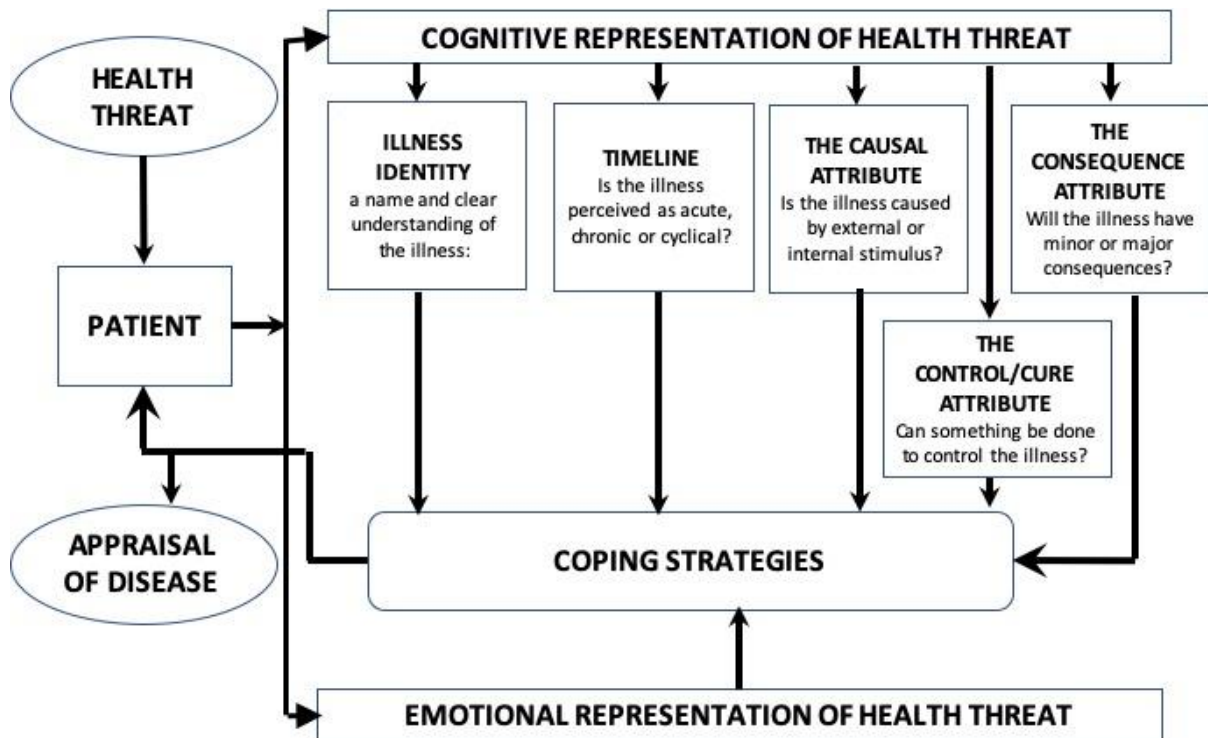


Figure 1 Schematic representation of a health threat in The Self-Regulation Model in FND.
Included and adapted for FND with kind permission from Parfeni, Nistor and Covic, 2013).

The SRM theorises that the perception of how long the illness will last is based on how the illness is represented cognitively. For example, depending on what is understood by the type of illness, there will be expectations from the person experiencing symptoms whether these will occur as a one-off experience, if they will last for a period of time or come and go at intervals. Responses to the cognitions around expected duration of symptoms are categorised as timeline in the SRM.

According to the SRM, potential causes in terms of whether internal or external factors are responsible for the illness developing will also be held in the cognitive representation, this is referred to as the causal attribute. Other factors contributing here include, the age of the individual, their prior experience of symptoms and the degree of success they have achieved trying to manage these. Experience from these factors also feed into the casual attribute in the model providing feedback as to whether anything can be done to control the symptoms (Leventhal, Halm, Horowitz, Leventhal, Ozakinci et al., 2004).

Beliefs held around management and recovery from illness are called the control/cure attribution in the SRM. These cognitions centre around expectations about the extent to which the illness can be managed or cured either by the body itself or with medication and/or other methods of intervention (Leventhal, Forster, Leventhal, Aldwin, Park, & Spiro, 2007). Expectations around what will happen as a result of having the illness is the consequence attribute within the SRM. Consequences of illness in the SRM can be categorised into four factors; physical function, personal, social and economic factors (Leventhal et al., 2007). In addition to the cognitive representation of the perceived health threat held in the SRM, there is a representation relating to how emotional responses to the illness are represented within the individual experiencing symptoms. Emotional reactions occur in parallel and link to how the threat is positioned cognitively (Leventhal et al., 2007). For example, symptoms persist despite efforts to control these and an emotional reaction of anxiety and low mood may occur as a result (Leventhal et al., 2007). The cognitive and emotional representations of illness in the SRM feed in to coping strategies which informs how the illness is viewed. This cycle of symptom appraisal in the SRM is a dynamic process with symptoms being re-evaluated when new information or changes are perceived. This can lead to changes in both how the illness is experienced physically and coped with emotionally (Horne & Weinman, 1998).

Previous research has identified that negative health beliefs can be detrimental to speed of recovery from illness (Donkin et al, 2005; Frostholt, Fink, Christensen, Toft, Oernboel, et al., 2005; Petrie & Weinman, 2006) and link to higher levels of healthcare utilisation (Frostholt et al., 2005). Negative health beliefs are common in those experiencing illnesses with a functional basis, such as chronic fatigue (Knoop, Prins, Moss-Morris & Bleeijenbergh, 2010), fibromyalgia (Glattacker, Opiz & Jackel, 2010) and irritable bowel syndrome (Rutter & Rutter, 2002). Taken together these studies indicate the utility of the SRM in providing a framework by which to consider the role of health beliefs in FND, however there are also limitations of the model to consider.

An obvious limitation of the SRM is it considers beliefs about the illness in isolation from other beliefs that an individual may hold about factors that may also influence recovery (Horne, 1997). For example beliefs about the appropriateness of the treatment on offer and the subsequent impact this has on patient engagement with the treatment process. This may be especially relevant for people with FND result in rejection of psychologically based treatment as they do not believe this is an appropriate treatment for their personal symptom profile (see Stone, Carson & Hallett, 2016). The attrition rates of people with FND from therapeutic interventions may be a demonstration of this. The SRM does not consider level of knowledge or the accuracy of pre-existing knowledge a patient holds about their condition. Often patients are not aware of what FND is and may have already established views that they have another diagnosis which can interfere with acceptance of an FND diagnosis (Stone et al., 2016). This review aims to establish if there are common themes of change within health beliefs after therapeutic intervention for those with FND using outcome measures based on the SRM.

Outcome Measures Based on the SRM

In order to evaluate health beliefs in illness using the framework of the SRM, three quantitative outcome measures have been developed the Illness Perceptions Questionnaire [IPQ (Weinman et al., 1996)]; the IPQ-Revised [IPQ-R; (Moss-Morris, Weinman, Petrie et al., 2002)] and the Brief Illness Perception Questionnaire [IPQ-B; (Broadbent, Petrie, Main & Weinman, 2006)]. Responses on the IPQ measure (Weinman et al., 1996) were initially divided into five scales assessing the components of cognitive representation of the illness: identity; timeline; control/cure; causal and consequences. A disadvantage of the original IPQ was the emotional representations of the illness were not considered in the instrument. The omission of the emotional representation was corrected when the IPQ was revised to include an item to represent this factor (the IPQ-Revised or IPQ-R) (Moss-Morris et al., 2002). Although this measure saw the inclusion of emotional representations held by the patient, the length of the questionnaire meant it took participants a significant amount of time to complete.

The third outcome measure based on the SRM was a briefer version of the IPQ-R, called IPQ-B (Broadbent et al., 2006). The IPQ-B consists of nine items rated on a scale from zero (minimum) to ten (maximum). Cognitive representations are measured by items one to five; perceptions such as effect on life (item one); length of time illness will last (item two); level of perceived control over illness (item three); beliefs around the effectiveness of treatment (item four); and beliefs about symptoms (item five). Emotional representations are explored by beliefs concern about illness (item six) and mood (item eight). The item on coherence measures how well the illness is understood (Item seven). Item nine is free text and offers space for the patient to rank what three things have caused their symptoms. A total score can be obtained which represents the level of threat perceived from the illness. To calculate the total score items three, four and seven are reverse coded

and added to the other items, a higher score is indicative the illness is perceived as more threatening (Broadbent et al., 2006). This measure is quick to complete and straight forward to interpret making it useful when working with people experiencing problems with their health.

Health Beliefs in FND

Negative health beliefs using the IPQ outcome measure have been found previously to be predictive of poorer health outcomes in FND (Sharpe et al., 2010). The research by Sharpe et al., (2010) used a prospective cohort design to collect data from Scottish Neurology clinics. Beliefs surrounding the permanence of symptoms and the magnitude of belief that psychological factors contributed to symptoms were measured using items from the IPQ (Weinman et al. 1996).

An additional factor predictive of outcome was receipt of health-related state payment benefits. It was suggested that receiving benefits may provide further validation to the symptoms and related levels of disability thereby contributing to the ongoing presence of FND (Sharpe et al., 2010). These findings contradicted the initial hypothesised outcomes would be dependent on the number of physical symptoms, the perceived level of disability and the severity of symptoms from anxiety/depression participants experienced (Sharpe et al., 2010).

This research utilised a design reflective of methods in clinical practice to explore health beliefs in an FND population, within the Scottish healthcare system. This result fits with evidence from other functional disorders where health beliefs negative in valence also resulted in poorer health outcomes (Glattacker et al, 2010; Knoop et al., 2010, Rutter & Rutter, 2002). There is a lack of treatment options available for those living with the condition to access (Division of Neuropsychology, 2013). Evidence to date has made it difficult to make a judgement regarding the clinical utility of introducing a therapeutic

intervention. For example, a study may measure changes perceived in physical symptoms (Mayor, Brown, Cock, House, Howlett et al., 2013); whereas others may explore changes in psychological experience (Conwill, Oakley, Evans & Cavanna, 2014) meaning it is difficult to capture overall benefits to patients taking part in therapeutic interventions.

Review Aims and Objective

In line with Sharpe et al., (2010), this review will focus on studies that mirror clinical practice and exclude research designs which do not e.g. Randomised Control Trial (RCT) designs. The decision to exclude RCT designs is bipartite. Firstly, the heterogeneous nature of the population with FND means that the controls may not reflect the same pattern of difficulties as the experimental group. Secondly, designs using outcome measures administered pre and post intervention are more reflective of outcomes collected in clinical practice. Exploring studies closer to clinical practice may be helpful in identifying potential patterns of change in the health beliefs of people with FND that take part in psychological interventions within clinical rather than research settings.

An additional facet of the study by Sharpe et al., (2010) is that health beliefs were quantified using measures based on the SRM. A limitation highlighted previously with developing an evidence base for effective treatment of FND was the heterogeneity of delivery methods and patient outcome measures in the research to date (Division of Neuropsychology, 2013). As a result, this review will also focus on studies utilising outcome measures based on the SRM namely the IPQ, IPQ-R and IPQ-B to consolidate evidence on one measure. As these measures are quantitative in nature no qualitative studies will be included in the review.

The overall objective of this review is to address a key gap in the current literature by exploring what elements of health beliefs as indexed by the SRM change for people diagnosed with FND after taking part in psychological interventions. Studies reporting outcomes based on the SRM for patients both pre and post engagement in a psychological intervention with and without a follow up will be included.

Method

Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines on how to undertake systematic reviews (Moher, Shamseer, Clarke, Ghera, Liberati et al., 2015) alongside guidance from the Centre for Reviews and Dissemination (CRD, 2009) informed the process of the current review (see Figure 2). Prior to conducting the full systematic review, preliminary literature searches were undertaken of the International Prospective Register of Systematic Reviews (PROSPERO) and the CRD to ensure that a review in this area had not been published previously or was currently registered as an active project. Scoping searches for potential review topics related to brief interventions for FND conducted prior to the current review included the efficacy of psychological interventions for FND and psychoeducation for FND. However, these topics had already been registered on the PROSPERO website resulting in a change of focus for the current systematic review topic.

Inclusion and Exclusion Criteria

Criteria for study inclusion and exclusion were developed using the 'PICOS' (Population, Intervention, Comparison, Outcome, Study design) model. (CRD, 2009).

Population

Adult patients (aged 16+) with a diagnosis of FND or a historical or alternative diagnostic equivalent were included e.g. conversion disorder; dissociative disorder; functional neurological symptom disorder; functional neurological symptom; functional

movement disorder; nonepileptic attacks; psychogenic symptoms; psychogenic non epileptic seizures (PNES); somatoform symptoms.

Intervention

Psychological interventions delivered either individually or in a group which included patient health beliefs as part of the outcomes for people with FND were included.

Interventions had to be based on well-established psychological principles such as cognitive behavioural therapy (CBT), acceptance and commitment therapy (ACT), psychoeducation, mindfulness etc.

Comparison

No control group was required for inclusion. Pre and post treatment measures where participants acted as own control.

Outcome

Studies that reported measures of health beliefs/illness perceptions based on the self-regulation model IPQ; IPQ-R; Brief-IPQ. Studies were excluded if qualitative or mixed methods analysis were used.

Study Design

Observational studies with quantitative methodology were included. Studies were excluded if they were published in non-peer reviewed media (e.g. conference abstracts) or when the full text article of the abstract was not available.

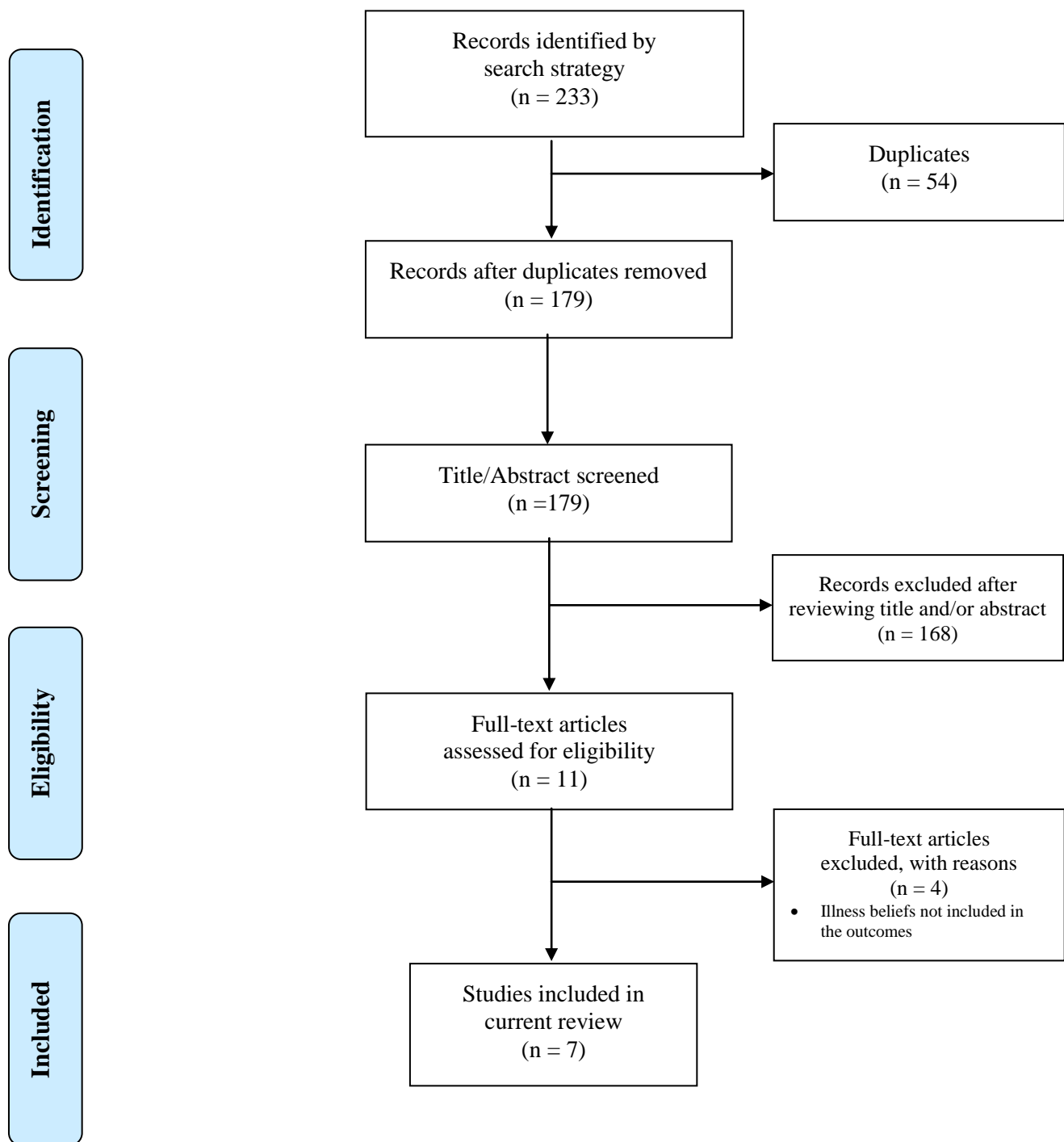


Figure 2 PRISMA Flow diagram

Literature Search Strategy

A systematic search was undertaken between January 1995 to January 2020: Ovid MEDLINE, PsycINFO, The Cochrane Register, EMBASE and Web of Science. The British Library Electronic Theses Online System (EThOS), and Google Scholar search engines were used to explore grey literature.

Search terms were based on previous research and current clinical documents were developed for capturing terms used for FND, health beliefs and psychological therapy in previous literature (see Table 1).

Table 1 Search strategy used in the systematic review

Area	Search Terms Used
FND	("conversion disorder*" or "dissociative disorder*" or "functional neurological disorder*" or "movement disorder*" or "Functional neurological symptom*" or "FND" or "PNES" or "nonepileptic" or "non epileptic" or "NEAD" or "somatoform" or "psychogenic symptom*").
Health Beliefs	("self-regulation" or "self-regulation" or "health belief*" or "illness perception*" or "health perception*" or "IPQ" or "common sense" or "patient attitude" or "illness cognition*" or "illness representation").
Psychological Intervention	((Cognitive behav* or CBT or psychological intervention* or psychological therap* or psychoeducation* or ACT or Acceptance) and Commitment*) or Psychother* or Brief Intervention* or Mindful* or Group Therapy* or Guided self* or Psychodynamic*).

The search strategy identified 233 studies. PICOS criteria were used to screen the titles and abstracts of studies to determine their suitability for inclusion. After screening, seven studies were identified as suitable for inclusion in the systematic review. All reference lists of the articles included in the review and articles citing the included papers were also searched to identify further papers. The protocol for this review was published by PROSPERO (see

https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42019158075h)

Data Extraction

PI (reviewer one) screened the title and abstract of studies using the review search strategy and compared against the inclusion/exclusion criteria. LM, a third-year trainee clinical psychologist (reviewer two) checked the data extracted for 4 papers randomly selected. Extracted data will be managed in a Microsoft Excel database prior to transferring to tables in the review document. References were managed by RefWorks to identify and remove duplicate articles.

Quality Assessment

The Newcastle Ottawa Scale assesses study quality using a star system over three categories: Selection (5-star maximum score), Comparability (maximum 2 stars) and Outcome (maximum 4 stars). The Newcastle Ottawa Scale is the recommended instrument for assessing non-randomised research (Julian, Higgins & Green, 2011) and has previously been used with outcome measures based on the SRM in systematic reviews in both physical health (Parfeni, Nistor & Covic, 2013) and a population with sudden onset neurological conditions (McAleese & Guzman, 2017).

For the purposes of this review the Newcastle Ottawa Scale was applied in the following way:

Selection / Representativeness (* per item)

1. Did the participants in the study represent people with FND in the community?
2. Were the number of participants justified, was the study sufficiently powered?
3. Were the differences between those taking part and not taking part explored and was the rate of recruitment satisfactory? (e.g. attrition rate between 5% and 20%)
4. Were the variables of interest in the study captured with validated outcome measures?

5. Was the version of the IPQ outcome measure used in line with the developers' recommendations?

Comparability (* per item)

6. Was there evidence that the study controlled for relevant predictors/risk factors/confounding variables (e.g. duration of symptoms?)

Outcome (* per item)

7. Was the assessment of outcome completed by independent blind assessment or unblinded assessment using objective validated laboratory methods *

OR Used non-standard or non-validated laboratory methods (e.g. double assessment of scored items *

8. Was follow up long enough for outcomes to occur with a clearly described and reported relevant statistical test? *

9. Were all subscales of the IPQ version used reported? *

In line with recommendation from the Centre for Reviews and Dissemination that quality assessment tools are adapted to reflect the aim of review. The Newcastle Ottawa Scale was amended with the inclusion of 2 items. The first item was added to the selection category to ascertain if the outcome measures based on the SRM are utilised in the study in line with the recommendations of the questionnaire developers. The second question was added to the outcome section to ensure the subscales of the IPQ were reported accurately and as recommended by the questionnaire developers (see Appendix 2).

High quality studies will be awarded 9-11 stars, Good Studies: 7-8 stars, Satisfactory Studies: 5-6 stars and Unsatisfactory Studies: 0 to 4 studies. PI (reviewer one) screened the title and abstract of studies using the review search strategy and compared against the inclusion/exclusion criteria. LM, a third-year trainee clinical psychologist (reviewer two) checked the data extracted from a subset of papers. It is recommended to minimise bias a minimum of two reviewers assessed the quality of the studies selected for inclusion (SIGN, 2015). Four of the included papers (57%) were selected at random and rated by LM (reviewer two), interrater reliability was found to be 80%. Discrepancies were discussed and resolved. The supervisory team were available to review independently if there were unresolved discrepancies between reviewers one and two.

Results

Study Characteristics

In line with the inclusion criteria of this review, all studies were prospective pre-post design, except for one which adopted a retrospective design (Saiffee, Kassavetis, Pareés, Kojovic, Fisher, et al., 2012). There were 3 studies that included a follow up period between 6 months (Goldstein, Deale, O'Malley, Toone & Mellers, 2004), 1 year (Demartini, Batla, Petrochilos, Fisher, Edwards, et al., 2014) and a median of 7 years (Saiffee et al., 2012).

The number of participants ranged from 9 – 66, with participants being predominantly female, the percentage of males participating ranged from 11% - 48%. Most studies (n=4) had participants only with non-epileptic attacks (Cope, Smith, King, & Agrawal, 2017; Goldstein et al., 2004; Williams, Howlett, Levita, & Reuber, 2018; Wiseman, Mousa, Howlett, & Reuber, 2016). The remaining 3 studies included people with other types of FND. All studies reported the duration participants had experienced symptoms which ranged from 3.6 years (Blake, Abilitt, Ruffmann, Morley, Williams et al., 2019) to 7.1 years (Cope et al., 2017). Employment rates were reported by all studies apart from one (Blake et al., 2019). The percentage of participants economically inactive ranged from 48% (Cope et al., 2017; Wiseman et al., 2016) to 87.5% (Goldstein et al., 2004).

Two studies delivered interventions within an inpatient rehabilitation setting (Demartini et al., 2014; Saiffee et al., 2012), the remaining five studies were outpatient based. Outpatient sessions ranged from three sessions lasting 90 minutes (Cope et al., 2017) to 20 sessions lasting 50 minutes (Williams et al., 2018). CBT was the most frequent modality used in the interventions with all studies drawing on this model except for Williams et al., (2018) who adopted brief augmented psychodynamic interpersonal therapy (BAPIT). Most of the therapy was delivered on an individual basis except for two studies which adopted a group format (Blake et al., 2019; Cope et al., 2017).

Illness perceptions were measured most frequently using the IPQ-B in three studies (Cope et al., 2017; Williams et al., 2018; Wiseman et al., 2016), two studies utilised the IPQ (Goldstein et al., 2004; Saifee et al., 2012) and two used the IPQ-R (Blake et al., 2019; Demartini et al., 2014).

Four out of seven studies used changes in symptoms as an outcome with all of these finding that participants symptoms improved post intervention (Cope et al., 2017; Goldstein et al., 2004; Saifee et al., 2012; Wiseman et al., 2016). Most studies (n=4) included measures evaluating the participants' experience of the intervention (Blake et al., 2019; Cope et al., 2017; Saifee et al., 2012; Wiseman et al., 2016). The characteristics of the included studies are summarised in Table 2.

Table 2 Characteristics of Included Studies

	Study	Method	Participants	Description of Intervention	Illness Perception Outcome Measure ¹	Other Outcome Measures ¹	Results
1	Blake et al., 2019 UK	Design: Prospective Pre-Post No comparative group Setting Outpatient	No of participants n = 9 Age M=44.1 (SD = 12.9) Gender 89% Female 11% Male Type of FND 3xPNES; 1 x Sensory; 6xFunctional Motor symptoms Duration of Symptoms M=3.6 years (SD = 3.6) Recruitment 10/16responded 9/10 took part pre-post Employment Not reported	Duration: 2-hour sessions Number of sessions 6 Sessions Group frequency not reported Modality: CBT based on existing manual Delivered By MDT Approach 1x Neurology; 1 x physiotherapy; 2 x psychology; 1 x nurse Staff experience: Clinical Psychologists/Neurologists Data Timepoints Pre-therapy; Post-therapy	Version: IPQ-R Administered: Entire questionnaire Interpretation Not all subscales reported	Self-Report HADS SF36 Study specific Patient Exp	Symptoms (self-report) Not reported Patient Experience 55% found helpful Reduced feelings of isolation More time for discussion Some felt no relevance to their symptoms and content too generic Outcomes Decrease in HADS score Increase in SF36 Phys and psych role limitations; social function; health change Decrease of 1.56 points on IPQ-R (pre 46.89; Post 45.33) Employment Not reported
2	Cope et al., (2017) UK	Design: Prospective Pre-Post No comparative group Setting Outpatient	No of participants n = 16 Age Age categories not mean reported 20% 18-25 32% 26-35 32% 36-45 46+ Gender 87.5% Female 12.5% Male Type of FND PNES Duration of Symptoms M=7.1 years (SD = 6.6) Recruitment Number invited not reported 19/25 took part pre-post Employment 48% Unemployed	Duration: 90-minute sessions Weekly Number of sessions 3 Sessions Modality: CBT Group Therapy Delivered By Clinical Psychologist Staff experience: Formal Clinical Psychology training Data Timepoints Pre-therapy; Post-therapy; 6months Follow Up	Version: IPQ-B Administered: Entire questionnaire Interpretation Not all subscales reported with stats	Self-Report Seizure Frequency DES-II (pre only) PHQ9 GAD-7 WSAS Study specific Patient Exp	Symptoms (self-report) 40% (7/18) Attack Free post intervention Patient Experience Felt less alone, reduced feelings of isolation. Valued space to share coping mechanisms. Prefer longer sessions and a follow up group. Outcomes Statistically significant improvements in ET7 and IPQB subscales relating to how long illness will continue Levels of concern and understanding of illness. Not related to illness duration Employment Not reported

Table 2 Continued

Study	Method	Participants	Description of Intervention	Illness Perception Outcome Measure ¹	Other Outcome Measures ¹	Results
3 Goldstein et al., 2014	Design: Prospective Longitudinal Follow Up 12 months No comparative group Setting Outpatient	No of participants n = 16 Age M=34.9 (SD = 13.4) Gender 87.5% Female 12.5% Male Type of FND PNES 18.75% (n=3) psychiatric comorbidities Duration of Symptoms M=3.6 years (SD = 3.6) Recruitment 20/20 responded 16/20 took part pre-post Employment Pre: 87.5% not employed (n=11 Unemployed; n=3 students) Pre: 95% health related benefits	Duration: Initial session 2 hours Additional sessions: 60 minutes Weekly or Fortnightly Number of sessions 12 Sessions Modality: CBT Individual Therapy Delivered By CBT Therapist (Nurse) Staff experience: Formal CBT training & supervision Data Timepoints Pre-therapy; Post-therapy; 6months Follow Up	Version: IPQ Administered: Entire questionnaire Interpretation Mean reduction Causes Physical (Pre m=1.91 SD =0.58; Post 1.56 SD = 0.49); Consequences (Pre m=3.71 SD =0.59; Post 3.32 SD =0.59) Control/Cure (Pre m=3.19; SD =0.52; Post 3.86 SD = 0.74)	Self-Report Seizure freq WSAS Fear Q'aire HADS MHLC	Symptoms (self-report) 81.25% either seizure free or 50% less seizure activity Patient Experience Not reported Outcomes Significant improvement from Pre – Post maintained at F-Up WSAS Avoidance decreased Fear Q'aire HADS Anxiety and depression IPQ (see previous column) Employment 31.25% (n=5; 4 previously unemployed; 1 student) moved into full-time employment (non-significant trend)
4 Demartini et al., (2014) UK	Design: Prospective Longitudinal Follow Up 12 months No comparative group Setting Inpatient Rehabilitation	No of participants n = 66 Age M=43.7 (SD = 70.2) Gender 70.2% Female 28.8% Male Type of FND 50.5% Functional Motor symptoms 21.2% PNES 18.8% Weakness 9.5% sensory Duration of Symptoms M=4.8 years (SD= 3.2) Recruitment Consecutive patients admitted between 2006-2008 26/32 responded 19/26 took part pre-post Employment Pre: 71% not employed Pre: 95% health related benefits	Duration: 4-week inpatient Number of sessions: Max of 20 (5 x Therapy over 4 weeks). Modality: CBT Individual Goal directed Delivered By MDT Approach CBT Therapist Occupational therapy Neurophysiotherapy Neuropsychiatry assessment and input Neurology assessment and input 1 x nurse Staff experience: Not reported Data Timepoints Admission; Discharge; Follow Up	Version: IPQ-R Administered: Entire questionnaire Interpretation Significant improvement IPQ-R reductions pre-post Subscales Acute/chronic; emotional representation; consequences Increased illness coherence;	Self-Report HoNos HADS Fear Q'aire PHQ15 CNSQ	Symptoms (self-report) Not reported Patient Experience Not measured Outcomes Significant increase Subscale: Illness coherence Significant increase pre-post & F-Up Subscale: Illness coherence Significant Reduction pre-post & F-Up Subscale: Emotional representation timeline (acute/chronic) illness coherence emotional representation between pre and post changes 12 month Follow up group Illness coherence and emotional representation Employment Not reported

Table 2 Continued

Study	Method	Participants	Description of Intervention	Illness Perception Outcome Measure ¹	Other Outcome Measures ¹	Results
5. Williams et al., (2018) UK	Design: Prospective Pre-Post (time between measures M=11.0 months; SD = 7.1) No comparative group Setting Outpatient	No of participants n = 44 Age M=41.5 (SD = 13.5) Gender 77.3% (n=34) Female 22.7% (n=10) Male Type of FND PNES Other – No information Duration of Symptoms 5.4 years (SD=10.8) Recruitment Consecutive patients 118/127 consented 72/118 completed Pre 44/72 completed Post Employment Pre: 63% economically inactive (Unemployed, on benefit, retired ill health or old age)	Duration: Initial session 2 hours Additional sessions: 50 minutes Number of sessions: Maximum = 20 Modality: Brief Augmented Psychodynamic Interpersonal Therapy (BAPIT) Individual Psychoeducation Delivered By 1 x psychotherapist Staff experience: Not reported Data Timepoints Pre-Post	Version: IPQ-B Administered: Entire questionnaire Interpretation Only overall illness threat score reported Mean reduction in IPQ-B reached significance at post (m=48.83; SD = 15.79) compared to Pre (M=55.51; SD = 11.84).	Self-Report EPS-25 PHQ-15 CORE-10 MHS PHS IPQ-Brief	Symptoms (self-report) Not reported Patient Experience Not reported Outcomes Emotion processing improved post treatment. Health related QoL improved IPQ-B Illness threat reduced significantly Employment Not reported
6. Wiseman et al., (2016) UK	Design: Prospective Pre-Post No comparative group Setting Outpatient 4 x centre multisite	No of participants n = 25 Age M=41.8 (SD = 18.1) Gender 52% Female 48% Male Type of FND PNES Duration of Symptoms 5.3 years (SD=9.1) Recruitment Consecutive patients 36/40 responded 29/36 attended 25/29 Completed 19/25 gave post-feedback Employment Pre: 13/25 52% in work	Duration: 1hour Psychoeducation Number of sessions: 4 x 1 hour Modality: CBT Individual Psychoeducation Delivered By 3 x Assistant Psychologists 1 x Occupational Therapist 6 x Specialist Epilepsy Nurses Staff experience: Limited Data Timepoints Pre-Post	Version: IPQ-B Administered: Entire questionnaire Interpretation Only overall illness threat score reported Mean reduction in IPQ-B reached significance at post (m=42; SD=21.3) compared to Pre (M = 54.5; SD = 23). Reported as improved understanding	Self-Report Seizure freq (1 month) WSAS CORE-OM NewQOL-6D EQ-5D Symptom attribution Study specific Patient experience	Symptoms (self-report) 6/25 seizure free for 1 month 6/25 reduced seizure activity Patient Experience 86% completed 96% reported found helpful 92% felt helped to control seizures 100% of these recommend 42% (8/19) agreeing to psychological constructs being causative post compared to 22% Pre (5/23) no stats for this Outcomes No significant relationship between duration of illness and outcomes CORE-10 significant decrease in psychological distress IPQ-B Illness threat reduced significantly Employment Not reported

Table 2 Continued

Study	Method	Participants	Description of Intervention	Illness Perception Outcome Measure ¹	Other Outcome Measures ¹	Results
7. Saifee et al., (2012) UK	Design: Retrospective Longitudinal Follow Up Setting Inpatient Rehabilitation	No of participants n = 19 Age M=47 (SD = 9.5) Gender 58% Female 42% Male Type of FND 68% Functional Motor symptoms 16% Weakness 16% Fatigue Duration of Symptoms >3years Recruitment Consecutive patients admitted between 2006-2008 26/32 responded 19/26 took part pre-post- Employment Pre: 16% in work Pre: 91% health related benefits	Duration: 4-week inpatient Number of sessions: 5 x Therapy Weekly. Modality: CBT Individual Goal directed Delivered By MDT Approach CBT Therapist Occupational therapy Neurophysiotherapy Neuropsychiatry assessment and input Neurology assessment and input 1 x nurse Staff experience: Not reported Data Timepoints Admission; Discharge; Follow Up Based on Self-help book Designed for routine care	Version: IPQ Administered: cause subscale only Interpretation Long term follow-up 27% agreed emotional factors causative 50% stress causative	Self-Report WSAS Study specific Patient experience	Symptoms (self-report) 58% improved 35% no change 7% deteriorated Patient Experience 65% found CBT helpful 58% reported found helpful 74% of these recommend 26% would not Outcomes No difference in which member of MDT most useful Correlation between participants reporting benefit and agreeing to psychological constructs being causative Employment 19% of patients in work at follow up (non-significant)

¹ **CNSQ** = Common Neurological Symptom Questionnaire; **CORE-10** = Clinical Outcomes in Routine Evaluation 10; **CORE-OM** = Clinical Outcomes in Routine Evaluation Outcome Measure; **COPM** = Canadian Occupational Performance Measure; **DES-II** = Dissociative Experiences Scale II; **EPS-25** = Emotional Processing Scale 25; **EQ-5D** = EuroQoL 5 dimensions; **ET7** = Revised Emotional Thermometer Scale; **GAD-7** = Generalised Anxiety Disorder Scale; **HADS** = Hospital Anxiety & Depression Scale; **HoNOS** = Health of the Nation Outcome Scale; **IPQ-B** = Illness Perception Questionnaire Brief; **IPQ** = Illness Perception Questionnaire; **IPQ-R** = Illness Perception Questionnaire Revised; **MHLC** = Multidimensional Health Locus of Control; **MHS** = Mental Health Summary; **NewQOL-6D** = QALY measure for Epilepsy; **PHQ-9** = Patient Health Questionnaire 9; **PHQ-15** = Patient Health Questionnaire 15; **PHS** = Physical Health Summary; **SF-36** = The Short Form 36; **WSAS** = Work and Social Adjustment Scale

Applying the Newcastle Ottawa Scale Quality Criteria

The quality assessment using the Newcastle Ottawa Scale suggested that four studies were categorised as satisfactory scoring between five and six stars on the quality rating (Cope et al., 2017; Demartini et al., 2014; Williams et al., 2018; Wiseman et al., 2016). Three studies were categorised as unsatisfactory (Blake et al., 2019 ; Goldstein 2014; Saiffee et al., 2012) scoring between two and four stars (See Table 3).

Selection

All studies scored between two and four stars in this section. Three studies met criteria for the representativeness of those with FND included in the studies and how it compared to people with FND in the community (Demartini et al., 2014; Saiffee et al., 2012; Williams et al., 2018;. One study recruited using multiple sources of referral, including neurology, psychiatry and GP's (Goldstein et al., 2004) however, this study and two others also used exclusion criteria which has implications for generalising the findings to the FND population. For example, two studies excluded participants with active psychiatric conditions (Blake et al., 2019; Cope et al., 2017), with two studies excluding patients with comorbid organic neurological conditions (Blake et al., 2019; Goldstein, 2014).

No formal power calculations were provided by any of the included studies. There were also no justifications regarding the number of participants that were recruited, resulting in no stars being awarded for any of the studies in this area (Blake et al., 2019; Cope et al., 2014; Demartini et al., 2014; Goldstein et al., 2004; Saiffee et al., 2012; Williams et al., 2018; Wiseman et al., 2016).

Table 3 Quality Rating for Studies Included in the Review

Quality Assessment Criteria		Acceptability (*)	1.Blake et al., (2019)	2.Cope et al., (2017)	3.Goldstein et al., (2014)	4.Demartini et al., (2014)	5.Williams et al., (2018)	6.Wiseman et al., (2016)	7.Saifee et al., (2012)
		Selection							
Representativeness of FND	Truly represents average person with FND in the community								
	OR Somewhat represents people with FND in the community		-	-	-	*	*	-	*
Sample size	Justified and satisfactory (including sample size calculation)		-	-	-	-	-	-	-
Non-respondents	Proportion of target sample recruited		-	-	*	*	*	*	*
Ascertainment of the exposure	Validated Outcome measures		*	*	*	*	*	*	*
IPQ/IPQ-R/IPQ-Brief administration	Consistent with developers' recommendations		*	*	*	*	*	*	-
		Comparability							
Confounding variables controlled for.	Data/Results controlled for relevant predictors/ risk factors/confounders (**)		-	**	-	-	-	**	-
	OR Used non-standard/validated methods (*)								
		Outcome							
Assessment of outcome	Independent blind assessment/record linkage (**)								
	OR Unblinded assessment using objective validated lab methods (**)		-	-	-	-	-	-	-
Follow Up	Was follow up long enough for outcomes to occur with a clearly described and reported relevant statistical test? *		-	*	*	*	*	*	*
IPQ/IPQ-R/IPQ-Brief reporting	Subscales reported in full		-	-	-	-	-	-	-
Overall Quality Rating¹ (Minimum = 0 Maximum =11)			2	5	4	5	5	6	4

¹Very good: 9-11 stars; Good 7-8 stars; Satisfactory 5-6 stars; Unsatisfactory 0 to 4 stars

The number of non-respondents in the reviewed studies was generally low, with two studies recruiting all the participants they invited (Demartini et al., 2014; Goldstein et al., 2004). Most of the others had between 7% - 18.75% not responding to the invitation to participate (Williams et al., 2018; Saifee et al., 2012; Wiseman et al., 2016). Out of the remaining studies, 37.5% of those invited did not respond (Blair et al., 2019) and one study did not report recruitment figures (Cope et al., 2017).

All studies utilised standardised outcome measures to collect data on the variables of interest (Blake et al., 2019; Cope et al., 2017; Demartini et al., 2014; Goldstein et al., 2004; Saifee et al., 2012; Wiseman et al., 2016; Williams et al., 2018).

Six studies appeared to administer their version of the IPQ in line with the developers' recommendations (Blake et al., 2019; Cope et al., 2014; Demartini et al., 2014; Goldstein et al., 2004; Wiseman et al., 2016; Williams et al., 2018). The remaining study administered only part of the IPQ (Saifee et al., 2012)

Comparability

All studies had predominantly female participants. This fits with the clinical profile of FND which is thought to be more common in females with prevalence ranging between 60-75% for those diagnosed (see Carson & Lehn, 2016). The mean age of participants in the included studies fitted in general with the age of average onset of FND reported to occur most often between the ages of 35–50 years old (see Carson & Lehn, 2016). However, FND is also common in men and can occur at any point over the lifespan (see Carson & Lehn, 2016). Only one study did not report a mean or median age, choosing instead to report frequency of age categories (Cope et al., 2017).

Diagnosis of FND in the participants was completed by neurologists in five studies (Blake et al., 2019; Cope et al., 2014; Demartini et al., 2014; Wiseman et al., 2016; Williams et al., 2018). One study (Goldstein et al., 2004) reported diagnosis was made by a

neuropsychiatrist, who utilised video-telemetry, thought to be the gold standard for diagnosis of non-epileptic seizures (Gedzelman & LaRoche, 2014). The final study had diagnosis confirmed by a multi-disciplinary team, which included neurologists (Saifee et al., 2012). It is considered unusual for FND to be present as a stand-alone set of symptoms. Common comorbidities often found in FND include pain, fatigue, weakness and sensory disturbances (Carson et al., 2000; Kim, Pakiam, & Lang, 1999; Koller, Lang, Vetere-Overfield, Findley, Cleaves et al., 1989).

Participants with active psychiatric conditions were excluded from three studies (Blake et al., 2019; Goldstein et al., 2004; Wiseman et al., 2016) however dissociation in non-seizures may be trauma related (Holmes, Brown, Mansell, Fearon, Hunter et al., 2005), therefore excluding on this criterion may be potentially omitting participants with predisposing factors to FND.

The presence of comorbid organic neurological conditions resulted in participants being excluded in two studies (Blake et al., 2019; Cope et al., 2017). Organic neurological conditions are also found more frequently in people with FND, occurring in approximately one in ten cases, which is higher probability than chance (Stone, et al., 2012). The exclusion of participants may risk selection bias in the sample. However, there also needs to be consideration regarding the safety of the participant and the ethical dilemma of including the patient in a research project whilst managing risks associated with poor mental health (e.g. risk of suicide and/or self-harm).

Outcome

All the included studies received no stars for outcome assessment. The studies did not include a control or comparison group therefore assessing the outcome by independent blind assessment was not possible in the delivery of the included intervention. The use of

other methods to minimise bias in the scoring of outcome assessments, such as double scoring of assessments, was not reported by any of the included studies, resulting in no stars on this category of the Newcastle Ottawa Scale. Finally, all the studies used outcomes potentially subject to bias as the measures were self-report.

Follow up periods varied between six months (Goldstein et al., 2004); one year (Demartini et al., 2014; Williams et al., 2018) and two years (Saifee et al., 2012). The sample sizes in one study led to it being so underpowered an analysis using inferential statistics was not possible (Blake et al., 2019). However, the remaining studies used appropriate statistical analysis and were all awarded a star on the Newcastle Ottawa Scale.

Assessment of the scoring and subsequent reporting of the included versions of the IPQ resulted in no studies reporting the subscales of their chosen measure in full. In addition, no study reported if Cronbach's alpha coefficients, in line with developer recommendations, were reported. There was no justification provided to support the omission of the reliability calculation or the omission of the items subscales in any of the included studies (Blake et al., 2019; Cope et al., 2017; Demartini et al., 2014; Goldstein et al., 2004; Saifee et al., 2012; Williams et al., 2018; Wiseman et al., 2016).

Narrative Synthesis

A meta-analysis was not considered for the current review due to the lack of homogeneity in the included study's methodology for variables such as the duration of sessions, the number of sessions and the variety of outcome measures used.

Two studies utilised a group format (Blake et al., 2019; Cope et al., 2017). However, the lack of inferential analysis of data in the second group study (Blake et al., 2019) does not allow these interventions to be compared.

Three of the included studies consisted of interventions only for participants with non-epileptic attacks. These were delivered in a group (Cope et al., 2017) and one to one format (Goldstein et al., 2004; Wiseman et al., 2016). Two of these studies utilised a cognitive behavioural model of treatment, using the IPQ-B as their index of health beliefs (Cope et al., 2017; Wiseman et al., 2016). The studies were similar in number and duration of sessions (3 x group sessions of 90 minutes; Cope et al., 2017; 4 x individual sessions of 1 hour); There were also differences in the level of experience of the facilitators with the individual session having less experienced staff compared to the group. Improvements were found in both studies post intervention for IPQ-B. Unfortunately, both studies reported their findings differently making comparison difficult. The group study reported individual subscales; three subscales measuring perception of how long the illness will continue, levels of concern and how well FND was understood showed significant improvement post intervention. However, the composite score representing level of threat perceived by the participant was not reported (Cope et al., 2017). In the one to one intervention, the level of threat perceived significantly reduced post intervention. However, individual subscales were not reported (Wiseman et al., 2016). Neither study reported significant changes in another shared outcome, the Work and Social Adjustment Scale (Mundt, Marks, Shear & Greist, 2002). Similar reductions in levels of participants' reporting being symptom free at the end

of the intervention were found in both studies (40%, Cope et al., 2017; 48%, Wiseman et al., 2016). Duration of symptoms was also unrelated to study outcomes in both interventions (Cope et al., 2017; Wiseman et al., 2016), however gender differences were not explored. Both studies incorporated participant feedback. This showed comparable levels of acceptability, indicating that the interventions were found to be helpful to those participating. An additional benefit of the group programme, highlighted in the feedback, was the potential that meeting others with the same diagnosis helped participants feel less isolated (Cope et al., 2017). This theme of feeling less isolated as a result of a group intervention was also reported in participant feedback for another study (Blake et al., 2019).

Individual therapy for Psychogenic Non-Epileptic Seizures using an alternative psychological model of brief augmented psychodynamic interpersonal therapy (BAPIT) also chose the IPQ-B in their outcomes (Williams et al., 2018). This sample was mixed FND, however initially data were originally analysed as two participant groups; those with PNES and those with other types of FND symptoms. As no significant differences were found between the subsets of participants, data were collapsed by the authors across the different types of FND, creating a sample of mixed FND pathology. Participants in the BAPIT intervention reported a similar level of perceived threat from their FND to the intervention that also utilised a cognitive behavioural approach on a one to one basis (Wiseman et al., 2016). Both studies reported significant reduction in the level of threat perceived from FND using the composite score from the IPQ-B and interpreted this effect as evidence of improvement in how the participants understood their FND (Williams et al., 2018; Wiseman et al., 2016). In contrast to the Wiseman et al (2016) study, Williams et al., (2018) did not include a measure of participant experience, therefore this cannot be compared across the studies. Furthermore, neither study included a follow up component to their research so the robustness of this effect cannot be determined.

Other studies using one to one with a mixed population were both undertaken by a multi-disciplinary team, within an inpatient rehabilitation setting (Demartini et al., 2014; Saifee et al., 2012). Both studies had a sample of participants with mixed FND pathology that took part in a 4-week inpatient intervention. One study found a statistically significant improvement at the end of treatment on the subscales of the IPQ-R. These were indexing of understanding of FND (illness coherence) level of distress experienced as a result of symptoms (emotional representation); symptoms as being less likely to be permanent (timeline acute/chronic) and felt to have less impact on their life (consequences) (Demartini et al., 2014). Attrition was high with 45% of those participating lost to follow up. However, for those who completed 12-month follow up differences were found post treatment and sustained for improvement in illness coherence and emotional representation (Demartini et al., 2014). In addition to health beliefs, 64% rated their FND symptoms as improved and 67% reported their general health was better or much better.

An earlier study with a different cohort of patients from the same inpatient programme conducted a retrospective follow up with a median of seven years (Saifee et al., 2012). Fifty eight percent of those who had participated in the intervention previously reported a sustained level of improvement compared to the symptoms they recalled when they took part (Saifee et al., 2012). The index of health beliefs was the IPQ in this study which revealed that at follow up participants believed that emotional state (27%) and 'stress' (50%) was causative. A correlational analysis found that those reporting psychological factors as potential causes appeared to benefit significantly more from the intervention (Saifee et al., 2012).

An additional study (Goldstein et al., 2004), also conducted a follow up analysis also using the IPQ but in an outpatient setting in a participant population with PNES. A significant difference was also found in this sample of participants for the belief that emotional factors had contributed more to their FND than physical factors, this difference remained significant at 6 months follow up (Goldstein et al., 2004). Other subscales of the IPQ reported in this study revealed that there was no significant change with regards the expected timeline of experiencing FND. However, there were significant improvements around the perceived impact FND would have on their life (consequences) and an increase in the amount of control perceived over symptoms (control/cure). All significant difference found post-treatment remained at the six month follow up.

Discussion

Treatment options for FND are in their infancy, with preliminary guidelines lacking clear direction for clinical services in how to operationalise the recommendations. There is a lack of consistent evidence for the best approach to treating FND with emerging guidelines lacking clarity on how to put their recommendations into practice. As a result, this review opted to focus on health beliefs which have been shown to be a valid predictor for recovery in FND previously (Sharpe et al., 2010). The beliefs identified were related to the permanence of symptoms and the magnitude to which psychological factors contributed to the symptoms being experienced. As Sharpe et al., (2010) utilised items from the IPQ (Weinman et al. 1996) which is based on the SRM, this review included treatment options utilising psychological therapy and including outcome measures based on the SRM.

As the guidelines for FND are to help embed treatment options in routine clinical practice, the results of the review are based on non-randomised studies that incorporated a health belief outcome measure based on the SRM as part of evaluating a psychological intervention for people with FND. The overall aim of this systematic review was to investigate if health beliefs for people diagnosed with FND change after taking part in psychological interventions.

All studies, that used inferential statistics, reported improvements in the health belief measures for participants taking part in psychological interventions. In terms of successful outcome, there appeared to be no added benefit to having one to one in contrast to group therapy. Indeed, group interventions may have added benefits in being more cost effective in terms of resources but also may indirectly address feelings of isolation in those with FND (Blake et al., 2019; Cope et al., 2017).

There appeared to be commonalities across subtypes of FND within the included study with regards health beliefs with no condition specific patterns being reported. Most included studies utilised a cognitive behavioural therapeutic approach, however, participants taking part in a BAPIT intervention showed similar reductions in the level of threat they perceived when compared to individuals taking part in a CBT based therapy (Williams et al., 2018; Wiseman et al., 2016).

The findings of the included research showed participants had more acceptance for psychological factors contributing to their FND symptoms post-intervention and at follow up (Demartini et al., 2014; Goldstein et al., 2004; Saifee et al., 2012). Participants also reported a greater understanding of FND across studies (Cope et al., 2017; Demartini et al., 2014; Saifee et al., 2017; Williams et al., 2018; Wiseman et al., 2016). Saifee et al., (2012) found that participants made better progress over time if they reported more acceptance of psychological factors contributing to symptoms, which fits with the findings from previous research (Sharpe et al., 2010).

Four out of seven studies were rated satisfactory in quality (Cope et al., 2017; Demartini et al., 2014; Williams et al., 2018; Wiseman et al., 2016). These studies tended to be robust in terms of recruitment ensuring a sample representative of those with FND in the community and showed smaller rates of attrition (Demartini et al., 2014; Williams et al., 2018). All studies utilised validated measures for their outcomes and the majority used their version of the IPQ in line with the developer's recommendations.

Areas of poorer quality related to controlling for potentially confounding variables, the use of self-report outcomes and the use of (or lack of reporting of) methods to minimise bias (e.g. having two people score outcome measures) were not used in the study method. Although all the IPQ health belief measures appeared to be administered correctly, there appeared to be a lack of consistency with the developer's recommendations of stating all

subscales on an individual basis and reporting the internal reliability within the sample, resulting in no studies earning stars in this area.

The area of research targeted by this review is novel and has evidence to suggest the area of health beliefs has predictive validity with patient outcome (Sharpe et al., 2010). In addition to a systematic search strategy, citation records were also consulted both for papers cited within the reference list of included papers and for papers citing the papers included. To address selection bias, a second reviewer (LM) also compared the selected studies against the review inclusion/exclusion criteria in addition to applying the quality criteria.

This review was limited as it contained only non-randomised studies. A further limitation was the authors of the included papers were not contacted to ascertain if the information regarding the non-reported subscales or scoring methods were available. In addition, only papers written in the English language were included potentially leading to a language bias within the review. How generalisable the results for this review are is difficult to decide based on these results alone. There were limited number of papers including health beliefs in their outcomes of therapy and those with small sample sizes may result in studies that were underpowered to detect effects on all measures (Blake et al., 2019; Cope et al., 2017; Goldstein et al., 2004; Saifee et al., 2012; Wiseman et al., 2016). Many studies were also cross-sectional with health beliefs as a secondary outcome rather than a main variable of interest (Blake et al., 2019; Cope et al., 2017; Wiseman et al., 2016)

Future research should consider the potential benefits of including an objective measure of health beliefs when conducting research in the efficacy of interventions for people with FND. Incorporating a follow up period where possible to explore the temporal aspect of these changes and determining if changes translate to meaningful improvements in function for participants would hold clinical utility. Although duration of symptoms was considered in

some analyses (Cope et al., 2017; Wiseman et al., 2016) these relationships were correlational within small samples.

It would be interesting to look at health beliefs and resulting outcomes for newly diagnosed participants taking part in psychological therapy to see if the timing of therapy has an impact on health beliefs. Exploring the impact of the experience level of the intervention facilitators on group outcomes could also be a useful study. The conclusion of this review is that there is tentative evidence to suggest psychological interventions have potential to change the maladaptive beliefs people hold about their FND. However, there needs to be more robust methods incorporated for the analysis and reporting of health belief outcomes. In addition, due to the lack of current treatment options, most participants had been living with their symptoms for some time with minimal support from healthcare providers. This lack of established treatment pathways may contribute to the feelings of isolation participants report as a result of living with FND. A more robust diagnosis process offering access to information around symptoms with the option of peer support may be beneficial for those being diagnosed with FND in the future.

References

- Ahmad, O., & Ahmad, K. E. (2016). Functional neurological disorders in outpatient practice: an Australian cohort. *Journal of Clinical Neuroscience*, 28, 93-96.
- Bateman, D. E., & Harrison, F. M. (2000). Psychiatric disorder in the neurology clinic. *Association of British Neurologists-University of Exeter*, 44.
- Benjamin, S., & Eminson, D. M. (1992). Abnormal illness behaviour: childhood experiences and long-term consequences. *International Review of Psychiatry*, 4, 55-69.
- Blake, J., Ablitt, A., Ruffmann, C., Morley, K., William, S., Moore, A., et al. (2019). Functional neurological disorder: Reflections on a pilot psychoeducational group in Hampshire. *The Neuropsychologist*, 8, 28-33.
- Broadbent, E., Petrie, K. J., Main, J., & Weinman, J. (2006). The brief illness perception questionnaire. *Journal of Psychosomatic Research*, 60(6), 631-637.
- Carson, A., & Lehn, A. (2016). Epidemiology. In M. Hallett, J. Stone & A. Carson (Eds.), *Handbook of clinical neurology* (pp. 147-160) Academic Press.
- Carson, A. J., Ringbauer, B., Stone, J., McKenzie, L., Warlow, C., & Sharpe, M. (2000). Do medically unexplained symptoms matter? A prospective cohort study of 300 new referrals to neurology outpatient clinics. *Journal of Neurology, Neurosurgery, and Psychiatry*, 68(2), 207-210.
- Centre for Dissemination and Reviews (CRD) (2009). Systematic Reviews: CRD's Guidance for undertaking Reviews in Health Care. York: Accessed December 2019
https://www.york.ac.uk/media/crd/Systematic_Reviews.pdf

- Cock, H. R., & Edwards, M. J. (2018). Functional neurological disorders: Acute presentations and management. *Clinical Medicine (London, England)*, 18(5), 414-417.
- Conwill, M., Oakley, L., Evans, K., & Cavanna, A. E. (2014). CBT-based group therapy intervention for nonepileptic attacks and other functional neurological symptoms: A pilot study. *Epilepsy & Behavior*, 34, 68-72.
- Cope, S. R., Smith, J. G., King, T., & Agrawal, N. (2017). Evaluation of a pilot innovative cognitive-behavioral therapy-based psychoeducation group treatment for functional non-epileptic attacks. *Epilepsy & Behavior*, 70, 238-244.
- Demartini, B., Batla, A., Petrochilos, P., Fisher, L., Edwards, M. J., & Joyce, E. (2014). Multidisciplinary treatment for functional neurological symptoms: A prospective study. *Journal of Neurology*, 261(12), 2370-2377.
- Division of Neuropsychology. (2013). *Management of patients with neurological medically unexplained symptoms within clinical services in NHS Scotland: The role of neuropsychology*. Scotland: BPS.
- Donkin, L., Ellis, C. J., Powell, R., Broadbent, E., Gamble, G., & Petrie, K. J. (2006). Illness perceptions predict reassurance following a negative exercise stress testing result. *Psychology and Health*, 21(4), 421-430.
- Edwards, M. J., Stone, J., & Lang, A. E. (2014). From psychogenic movement disorder to functional movement disorder: It's time to change the name. *Movement Disorders*, 29(7), 849-852.

- Frostholm, L., Fink, P., Christensen, K. S., Toft, T., Oernboel, E., Olesen, F., & Weinman, J. (2005). The patients' illness perceptions and the use of primary health care. *Psychosomatic medicine*, 67(6), 997-1005.
- Gedzelman, E. R., & LaRoche, S. M. (2014). Long-term video EEG monitoring for diagnosis of psychogenic nonepileptic seizures. *Neuropsychiatric disease and treatment*, 10, 1979-1986.
- Glattacker, M., Opitz, U., & Jäckel, W. (2010). Illness representations in women with fibromyalgia. *British Journal of Health Psychology*, 15(2), 367-387.
- Goldstein, L. H., Deale, A. C., O'Malley, S. J. M., Toone, B. K., & Mellers, J. D. (2004). An evaluation of cognitive behavioral therapy as a treatment for dissociative seizures: A pilot study. *Cognitive and Behavioral Neurology*, 17(1), 41-49.
- Healthcare Improvement Scotland (HIS). (2012). Stepped care for functional neurological symptoms: A new approach to improving outcomes for a common neurological problem in Scotland. *Edinburgh, National Health Service, Healthcare Improvement Scotland*.
- Holmes, E. A., Brown, R. J., Mansell, W., Fearon, R. P., Hunter, E. C., Frasquilho, F., et al. (2005). Are there two qualitatively distinct forms of dissociation? A review and some clinical implications. *Clinical Psychology Review*, 25(1), 1-23.
- Horne, R. (1997). Representations of medication and treatment: advances in theory and measurement. *Perceptions of health and illness*, 155-188.
- Horne, R., & Weinman, J. (1998). Predicting treatment adherence. *Adherence to Treatment in Medical Conditions*, Edited by Lynn Myers and Kenny Midence, 25-50.

- Hotopf, M., Mayou, R., Wadsworth, M., & Wessely, S. (1999). Childhood risk factors for adults with medically unexplained symptoms: results from a national birth cohort study. *American Journal of Psychiatry*, 156, 1796-1800.
- Julian, P. T., Higgins, S. G., & Green, S. (2011). Cochrane handbook for systematic reviews of interventions. The Cochrane Collaboration.
- Kim, Y. J., Pakiam, A. S., & Lang, A. E. (1999). Historical and clinical features of psychogenic tremor: A review of 70 cases. *Canadian Journal of Neurological Sciences*, 26(3), 190-195.
- Knoop, H., Prins, J. B., Moss-Morris, R., & Bleijenberg, G. (2010). The central role of cognitive processes in the perpetuation of chronic fatigue syndrome. *Journal of Psychosomatic Research*, 68(5), 489-494.
- Koller, W., Lang, A., Vetere-Overfield, B., Findley, L., Cleaves, L., Factor, S., et al. (1989). Psychogenic tremors. *Neurology*, 39(8), 1094-1099.
- Leventhal, H., Forster, R., Leventhal, E., Aldwin, C., Park, C., & Spiro, A. (2007). Self-regulation of health threats, affect, and the self. *Handbook of Health Psychology and Aging*, 341-366.
- Leventhal, H., Nerenz, D., & Steele, D. (1984). Illness representations and coping with health threats in Baum A, Taylor SE, singer JE, editors. handbook of psychology and health. volume IV: Social psychological aspects of health.
- Leventhal, H., Halm, E., Horowitz, C., Leventhal, E. A., Ozakinci, G., Sutton, S., et al. (2004). The SAGE handbook of health psychology.

- Leventhal, H., Meyer, D., & Nerenz, D. (1980). The common-sense representation of illness danger. *Contributions to Medical Psychology*, 2, 7-30.
- Mayor, R., Brown, R. J., Cock, H., House, A., Howlett, S., Smith, P., et al. (2013). A feasibility study of a brief psycho-educational intervention for psychogenic nonepileptic seizures. *Seizure*, 22(9), 760-765.
- McAleese, N., & Guzman, A. (2017). Investigation of the lived experiences and illness perceptions of adults with sudden onset neurological conditions.
- Moher, D., Shamseer, L., Clarke, M., Ghersi, D., Liberati, A., Petticrew, M., ... & Stewart, L. A. (2015). Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015 statement. *Systematic reviews*, 4(1), 1.
- Monzoni, C. M., Duncan, R., Grünewald, R., & Reuber, M. (2011). How do neurologists discuss functional symptoms with their patients: a conversation analytic study. *Journal of psychosomatic research*, 71(6), 377-383.
- Moss-Morris, R., Weinman, J., Petrie, K., Horne, R., Cameron, L., & Buick, D. (2002). The revised illness perception questionnaire (IPQ-R). *Psychology and Health*, 17(1), 1-16.
- Mundt, J. C., Marks, I. M., Shear, M. K., & Greist, J. M. (2002). The work and social adjustment scale: A simple measure of impairment in functioning. *The British Journal of Psychiatry*, 180(5), 461-464.
- Parfeni, M., Nistor, I., & Covic, A. (2013). A systematic review regarding the association of illness perception and survival among end-stage renal disease patients. *Nephrology Dialysis Transplantation*, 28(10), 2407-2414.

- Petrie, K. J., & Weinman, J. (2006). Why illness perceptions matter. *Clinical Medicine (London, England)*, 6(6), 536-539.
- Rommelfanger, K. S., Factor, S. A., LaRoche, S., Rosen, P., Young, R., & Rapaport, M. H. (2017). Disentangling stigma from functional neurological disorders: Conference report and roadmap for the future. *Frontiers in Neurology*, 8, 106.
- Rutter, C. L., & Rutter, D. R. (2002). Illness representation, coping and outcome in irritable bowel syndrome (IBS). *British Journal of Health Psychology*, 7(4), 377-391.
- Saifee, T., Kassavetis, P., Pareés, I., Kojovic, M., Fisher, L., Morton, L., et al. (2012). Inpatient treatment of functional motor symptoms: A long-term follow-up study. *Journal of Neurology*, 259(9), 1958-1963.
- Sharpe, M., Stone, J., Hibberd, C., Warlow, C., Duncan, R., Coleman, R., et al. (2010). Neurology out-patients with symptoms unexplained by disease: Illness beliefs and financial benefits predict 1-year outcome. *Psychological Medicine*, 40(4), 689-698.
- Stewart, D. E. (1990). The changing faces of somatization. *Psychosomatics*, 31, 153-158.
- Stone, J., Binzer, M., & Sharpe, M. (2004). Illness beliefs and locus of control: a comparison of patients with pseudoseizures and epilepsy. *Journal of psychosomatic research*, 57, 541-547.
- Stone, J., Carson, A., Duncan, R., Roberts, R., Warlow, C., et al., (2010). Who is referred to neurology clinics?—the diagnoses made in 3781 new patients. *Clinical neurology and neurosurgery*, 112, 747-751.

- Stone, J., Carson, A., Duncan, R., Roberts, R., Coleman, R., Warlow, C., et al. (2012). Which neurological diseases are most likely to be associated with “symptoms unexplained by organic disease”. *Journal of Neurology*, 259(1), 33-38.
- Stone, J., Carson, A., & Hallett, M. (2016). Explanation as treatment for functional neurologic disorders. In *Handbook of clinical neurology* (Vol. 139, pp. 543-553). Elsevier.
- Stone, J., Carson, A., & Sharpe, M. (2005). Functional symptoms and signs in neurology: Assessment and diagnosis. *Journal of Neurology, Neurosurgery, and Psychiatry*, 76 Suppl 1, i2-12.
- Stone, J., & Sharpe, M. (2002). Amnesia for childhood in patients with unexplained neurological symptoms. *Journal of Neurology, Neurosurgery & Psychiatry*, 72(3), 416-417.
- Stone, J., Sharpe, M., Deary, I., & Warlow, C. (2004). Functional paresis--paradoxes in illness beliefs and disability in 107 subjects. *Journal of Neurology, Neurosurgery and Psychiatry*, 75, 519-520.
- Stone, J., Wojcik, W., Durrance, D., Carson, A., Lewis, S., MacKenzie, L., et al. (2002). What should we say to patients with symptoms unexplained by disease? the "number needed to offend". *BMJ (Clinical Research Ed.)*, 325(7378), 1449-1450.
- Weinman, J., Petrie, K. J., Moss-Morris, R., & Horne, R. (1996). The illness perception questionnaire: A new method for assessing the cognitive representation of illness. *Psychology and Health*, 11(3), 431-445.

Williams, I. A., Howlett, S., Levita, L., & Reuber, M. (2018). Changes in emotion processing following brief augmented psychodynamic interpersonal therapy for functional neurological symptoms. *Behavioural and Cognitive Psychotherapy*, 46(3), 350-366.

Wiseman, H., Mousa, S., Howlett, S., & Reuber, M. (2016). A multicenter evaluation of a brief manualized psychoeducation intervention for psychogenic nonepileptic seizures delivered by health professionals with limited experience in psychological treatment. *Epilepsy & Behavior*, 63, 50-56.

Chapter 1: Appendices

Appendix 1 Submission guidelines for Neuropsychology review



Submission guidelines

Contents

Instructions for Authors

Types of papers

Review, Editorial, Commentary

Manuscript Standards

Manuscript Submission

Manuscripts submitted to Neuropsychology Review should conform to the style of the American Psychological Association Publication Manual (6th edition: 2010). Neuropsychology Review is an EQUATOR adopter. The EQUATOR network represents a collaboration of researchers and journal editors who aspire to improve accuracy and transparency in research by promoting better reporting standards. Because Neuropsychology Review publishes review articles, the EQUATOR elements most relevant are the PRISMA guidelines for preparation and reporting of systematic reviews and meta-analyses (<http://www.equator-network.org/reporting-guidelines/prisma/>).

While narrative reviews will still be considered for publication when appropriate, Neuropsychology Review encourages publication of systematic reviews of treatment, intervention and diagnostic validity studies as well as systematic reviews of research relating to scientific questions in all aspects of clinical neuropsychology and behavioral neuroscience. Systematic reviews are enhanced by inclusion of a carefully conducted meta-analysis whenever appropriate. Authors of systematic reviews and meta-analyses submitted to Neuropsychology Review should prepare their manuscripts according to the PRISMA guidelines and include a PRISMA checklist (<http://prisma-statement.org/PRISMA Statement/Checklist.aspx>) with manuscript submission. When completing the checklist, authors should consider whether their manuscript requires editing to address all of the reporting requirements.

Neuropsychology Review discourages use of numerical rating scales that assign a single number to rank the quality of studies included in the review. Instead authors should separately rate or classify individual study quality and risk of bias using established criteria such as those included in the critical appraisal checklists (e.g., randomized controlled trials or diagnostic validity studies (<http://www.cebm.net/critical-appraisal/>)). For treatment and intervention studies key risk-of-bias criteria include, but may not be limited to, adequacy of randomization, pre-treatment equality of groups, blinding of patients, therapist or person undertaking outcome evaluation, adequacy of follow-up and objectivity in outcome measurement. For diagnostic validity studies, risk-of-bias criteria include representativeness of sampling, full information on the test-to-be-evaluated (the index test) and diagnostic group status (the reference standard) and independent, blinded acquisition of reference and index test information. Other risk of bias criteria may be important in some contexts including commercial or other conflict of interest.

Prior to undertaking their systematic review, authors are encouraged to read the PRISMA Explanation and Elaboration paper (<http://www.ncbi.nlm.nih.gov/pubmed/19621070>). For authors not familiar with preparation of systematic reviews or the PRISMA guidelines, there are extensive information resources available on the PRISMA website (<http://www.prisma-statement.org/>).

Authors are encouraged to register their systematic review protocol early in the review process (e.g., PROSPERO), and use the PRISMA extension specifically written for reporting a systematic review protocol (i.e., PRISMA-P (<http://www.equator-network.org/reporting-guidelines/prisma-protocols/>)).

Authors of narrative reviews that are not based on systematic literature searching should justify in their cover letter and in the body of their manuscript why a systematic review was not feasible or appropriate. Likewise, authors of systematic reviews without meta-analysis should explain in their cover letter and in the body of their

manuscript why meta-analysis was not considered appropriate (e.g., reviewed studies were not of sufficient quality).

Authors should avoid use of non-standard abbreviations.

Manuscript Submission

Manuscript Submission

Submission of a manuscript implies: that the work described has not been published before; that it is not under consideration for publication anywhere else; that its publication has been approved by all co-authors, if any, as well as by the responsible authorities – tacitly or explicitly – at the institute where the work has been carried out. The publisher will not be held legally responsible should there be any claims for compensation.

Permissions

Authors wishing to include figures, tables, or text passages that have already been published elsewhere are required to obtain permission from the copyright owner(s) for both the print and online format and to include evidence that such permission has been granted when submitting their papers. Any material received without such evidence will be assumed to originate from the authors.

Online Submission

Please follow the hyperlink “Submit online” on the right and upload all of your manuscript files following the instructions given on the screen.

Please ensure you provide all relevant editable source files. Failing to submit these source files might cause unnecessary delays in the review and production process.

Title Page

The title page should include:

The name(s) of the author(s)

A concise and informative title

The affiliation(s) of the author(s), i.e. institution, (department), city, (state), country

A clear indication and an active e-mail address of the corresponding author

If address information is provided with the affiliation(s) it will also be published.

For authors that are (temporarily) unaffiliated we will only capture their city and country of residence, not their e-mail address unless specifically requested.

Abstract

Please provide an abstract of 150 to 250 words. The abstract should not contain any undefined abbreviations or unspecified references.

Keywords

Please provide 4 to 6 keywords which can be used for indexing purposes.

Text

Text Formatting

Manuscripts should be submitted in Word.

Use a normal, plain font (e.g., 10-point Times Roman) for text.

Use italics for emphasis.

Use the automatic page numbering function to number the pages.

Do not use field functions.

Use tab stops or other commands for indents, not the space bar.

Use the table function, not spreadsheets, to make tables.

Use the equation editor or MathType for equations.

Save your file in docx format (Word 2007 or higher) or doc format (older Word versions).

Headings

Please use no more than three levels of displayed headings.

Abbreviations

Abbreviations should be defined at first mention and used consistently thereafter.

Footnotes

Footnotes can be used to give additional information, which may include the citation of a reference included in the reference list. They should not consist solely of a reference citation, and they should never include the bibliographic details of a reference. They should also not contain any figures or tables.

Footnotes to the text are numbered consecutively; those to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data). Footnotes to the title or the authors of the article are not given reference symbols.

Always use footnotes instead of endnotes.

Acknowledgments

Acknowledgments of people, grants, funds, etc. should be placed in a separate section on the title page. The names of funding organizations should be written in full.

References

Citation

Cite references in the text by name and year in parentheses. Some examples:

Negotiation research spans many disciplines (Thompson 1990).

This result was later contradicted by Becker and Seligman (1996).

This effect has been widely studied (Abbott 1991; Barakat et al. 1995; Kelso and Smith 1998; Medvec et al. 1999).

Ideally, the names of six authors should be given before et al. (assuming there are six or more), but names will not be deleted if more than six have been provided.

Reference list

The list of references should only include works that are cited in the text and that have been published or accepted for publication. Personal communications and unpublished works should only be mentioned in the text. Do not use footnotes or endnotes as a substitute for a reference list.

Reference list entries should be alphabetized by the last names of the first author of each work.

Journal names and book titles should be *italicized*.

Journal article Harris, M., Karper, E., Stacks, G., Hoffman, D., DeNiro, R., Cruz, P., et al. (2001). Writing labs and the Hollywood connection. *Journal of Film Writing*, 44(3), 213–245.

Article by DOI Slifka, M. K., & Whitton, J. L. (2000) Clinical implications of dysregulated cytokine production. *Journal of Molecular Medicine*, <https://doi.org/10.1007/s001090000086>

Book Calfee, R. C., & Valencia, R. R. (1991). *APA guide to preparing manuscripts for journal publication*. Washington, DC: American Psychological Association.

Book chapter O'Neil, J. M., & Egan, J. (1992). Men's and women's gender role journeys: Metaphor for healing, transition, and transformation. In B. R. Wainrib (Ed.), *Gender issues across the life cycle* (pp. 107–123). New York: Springer.

Online document Abou-Allaban, Y., Dell, M. L., Greenberg, W., Lomax, J., Peteet, J., Torres, M., & Cowell, V. (2006). Religious/spiritual commitments and psychiatric practice. Resource document. American Psychiatric Association.

http://www.psych.org/edu/other_res/lib_archives/archives/200604.pdf. Accessed 25 June 2007.

For authors using EndNote, Springer provides an output style that supports the formatting of in-text citations and reference list.

Tables

All tables are to be numbered using Arabic numerals.

Tables should always be cited in text in consecutive numerical order.

For each table, please supply a table caption (title) explaining the components of the table.

Identify any previously published material by giving the original source in the form of a reference at the end of the table caption.

Footnotes to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data) and included beneath the table body.

Figure Numbering

All figures are to be numbered using Arabic numerals.

Figures should always be cited in text in consecutive numerical order.

Figure parts should be denoted by lowercase letters (a, b, c, etc.).

If an appendix appears in your article and it contains one or more figures, continue the consecutive numbering of the main text. Do not number the appendix figures, "A1, A2, A3, etc." Figures in online appendices (Electronic Supplementary Material) should, however, be numbered separately.

Figure Captions

Each figure should have a concise caption describing accurately what the figure depicts.

Include the captions in the text file of the manuscript, not in the figure file.

Figure captions begin with the term **Fig.** in bold type, followed by the figure number, also in bold type.

No punctuation is to be included after the number, nor is any punctuation to be placed at the end of the caption.

Identify all elements found in the figure in the figure caption; and use boxes, circles, etc., as coordinate points in graphs.

Identify previously published material by giving the original source in the form of a reference citation at the end of the figure caption.

Figure Placement and Size

Figures should be submitted separately from the text, if possible.

When preparing your figures, size figures to fit in the column width.

For large-sized journals the figures should be 84 mm (for double-column text areas), or 174 mm (for single-column text areas) wide and not higher than 234 mm.

For small-sized journals, the figures should be 119 mm wide and not higher than 195 mm.

Permissions

If you include figures that have already been published elsewhere, you must obtain permission from the copyright owner(s) for both the print and online format. Please be aware that some publishers do not grant electronic rights for free and that Springer will not be able to refund any costs that may have occurred to receive these permissions. In such cases, material from other sources should be used.

Accessibility

In order to give people of all abilities and disabilities access to the content of your figures, please make sure that

All figures have descriptive captions (blind users could then use a text-to-speech software or a text-to-Braille hardware)

Patterns are used instead of or in addition to colors for conveying information (colorblind users would then be able to distinguish the visual elements)

Any figure lettering has a contrast ratio of at least 4.5:1

Ethical Responsibilities of Authors

This journal is committed to upholding the integrity of the scientific record. As a member of the Committee on Publication Ethics (COPE) the journal will follow the COPE guidelines on how to deal with potential acts of misconduct.

Authors should refrain from misrepresenting research results which could damage the trust in the journal, the professionalism of scientific authorship, and ultimately the entire scientific endeavour. Maintaining integrity of the research and its presentation is helped by following the rules of good scientific practice, which include*:

The manuscript should not be submitted to more than one journal for simultaneous consideration.

The submitted work should be original and should not have been published elsewhere in any form or language (partially or in full), unless the new work concerns an expansion of previous work. (Please provide transparency on the re-use of material to avoid the concerns about text-recycling ('self-plagiarism').

A single study should not be split up into several parts to increase the quantity of submissions and submitted to various journals or to one journal over time (i.e. 'salami-slicing/publishing').

Concurrent or secondary publication is sometimes justifiable, provided certain conditions are met. Examples include: translations or a manuscript that is intended for a different group of readers.

Results should be presented clearly, honestly, and without fabrication, falsification or inappropriate data manipulation (including image based manipulation). Authors should adhere to discipline-specific rules for acquiring, selecting and processing data.

No data, text, or theories by others are presented as if they were the author's own ('plagiarism'). Proper acknowledgements to other works must be given (this includes material that is closely copied (near verbatim), summarized and/or paraphrased), quotation marks (to indicate words taken from another source) are used for verbatim copying of material, and permissions secured for material that is copyrighted.

Important note: the journal may use software to screen for plagiarism.

Authors should make sure they have permissions for the use of software, questionnaires/(web) surveys and scales in their studies (if appropriate).

Authors should avoid untrue statements about an entity (who can be an individual person or a company) or descriptions of their behavior or actions that could potentially be seen as personal attacks or allegations about that person.

Research that may be misapplied to pose a threat to public health or national security should be clearly identified in the manuscript (e.g. dual use of research). Examples include creation of harmful consequences of biological agents or toxins, disruption of immunity of vaccines, unusual hazards in the use of chemicals, weaponization of research/technology (amongst others).

Authors are strongly advised to ensure the author group, the Corresponding Author, and the order of authors are all correct at submission. Adding and/or deleting authors during the revision stages is generally not permitted, but in some cases may be warranted. Reasons for changes in authorship should be explained in detail. Please note that changes to authorship cannot be made after acceptance of a manuscript.

*All of the above are guidelines and authors need to make sure to respect third parties rights such as copyright and/or moral rights.

Upon request authors should be prepared to send relevant documentation or data in order to verify the validity of the results presented. This could be in the form of raw data, samples, records, etc. Sensitive information in the form of confidential or proprietary data is excluded. If there is suspicion of misbehavior or alleged fraud the Journal and/or Publisher will carry out an investigation following COPE guidelines. If, after investigation, there are valid concerns, the author(s) concerned will be contacted under their given e-mail address and given an opportunity to address the issue. Depending on the situation, this may result in the Journal's and/or Publisher's implementation of the following measures, including, but not limited to:

If the manuscript is still under consideration, it may be rejected and returned to the author. If the article has already been published online, depending on the nature and severity of the infraction:

- an erratum/correction may be placed with the article
- an expression of concern may be placed with the article
- or in severe cases retraction of the article may occur.

The reason will be given in the published erratum/correction, expression of concern or retraction note. Please note that retraction means that the article is maintained on the platform, watermarked "retracted" and the explanation for the retraction is provided in a note linked to the watermarked article.

The author's institution may be informed

Appendix 2: Adapted Newcastle-Ottawa Scale

Selection: (Maximum 5 star)

1. Representativeness of the sample:
 - a. Truly representative of people with FND. * (all subjects or random sampling)
 - b. Somewhat representative of the average in the target group. * (non-random sampling)
 - c. Selected group of users/convenience sample.
 - d. No description of the derivation of the included subjects.
2. Sample size:
 - a. Justified and satisfactory (including sample size calculation). *
 - b. Not justified.
 - c. No information provided
3. Non-respondents:
 - a. Proportion of target sample recruited attains pre-specified target or basic summary of non-respondent characteristics in sampling frame recorded. *
 - b. Unsatisfactory recruitment rate, no summary data on non-respondents.
 - c. No information provided
4. Ascertainment of the exposure:
 - a. Validated outcome measures. *
 - b. Description of non-validated outcome measures.
 - c. No details of outcome measures
5. Version of IPQ outcome measure used in line with developer's recommendations:
 - a. Yes. *
 - b. No

Comparability: (Maximum 2 stars)

1. Comparability of subjects in different outcome groups on the basis of design or analysis. Confounding factors controlled.
 - a. Data/ results adjusted for relevant predictors/risk factors/confounders e.g. age, sex, time since vaccination, etc. **
 - b. Data/results not adjusted for all relevant confounders/risk factors/information not provided.

Outcome: (Maximum 4 stars)

1. Assessment of outcome:
 - a. Independent blind assessment using objective validated laboratory methods. **
 - b. Unblinded assessment using objective validated laboratory methods. **
 - c. Used non-standard or non-validated laboratory methods with gold standard.
*
 - d. No description/non-standard laboratory methods used.
2. Follow Up:
 - a. yes if 3 months or longer*
 - b. No if no follow up or < 3 months.
3. Health Belief Reporting:
 - a. Version of IPQ outcome dimensions reported in full *
 - b. Version of IPQ Dimension partially reported
 - c. Results not reported

Study Rating:

Very Good Studies: 8-11 Stars

Good Studies: 7-8 points

Satisfactory Studies: 5-6 points

Unsatisfactory Studies: 0 to 4 points

In line with recommendation from the Centre for Reviews and Dissemination that quality assessment tools are adapted to reflect the aims of systematic reviews, this scale has been modified from the Newcastle-Ottawa Quality Assessment Scale cross-sectional studies to reflect variable of interest in this review.

A brief group intervention to support people with Functional Neurological Disorder in the Acute Sector: A feasibility study.

*Pauline Insch ^{1,2} Clara Calia ² and Fiona Summers ¹

¹ Department of Clinical Neuropsychology, NHS Grampian. Aberdeen, Scotland

² School of Health and Social Science, The University of Edinburgh, Scotland

*Corresponding author

E:Mail: S17947031@sms.ed.ac.uk

For submission to *Neuropsychology*

Word Count (excluding references and appendices): 17, 054

Empirical Paper Abstract

Introduction

Lack of treatment options can impact negatively on the quality of life of those with Functional Neurological Disorders (FND). This study aimed to evaluate the efficacy, feasibility and acceptability of a brief intervention for routine care designed to address a gap in current service provision within NHS Grampian.

Method

A two session intervention session was developed. Outcomes collected included cognitive function, mood, quality of life and health beliefs; Healthcare utilisation and associated costs and patient experiential data. Twenty nine participants (20 female) completed baseline measures, 16 participants (10 female) attended the first group, differences between attenders and non-attenders were explored. Eleven participants completed outcome measures over the four data timepoints.

Results

Psychological Measures Those attending the intervention rated their physical health limitations as significantly less in addition to higher levels of perceived personal control than non-attenders. Those participating in the intervention reported significantly reduced fatigue at follow up. Understanding of FND improved post intervention and this increase in knowledge was maintained at follow up.

Healthcare Utilisation and Costs: No differences between attenders and non-attenders. 63% of completers showed improved health status at follow-up on health-related quality of life.

Patient Experience Measures: Participants reported the brief intervention met their expectations, subjective levels of self-report knowledge about FND increased significantly from pre to post intervention.

Discussion

The brief intervention showed promising results in the exploratory study in terms of efficacy, feasibility and acceptability. Further research is required to ascertain if these findings are maintained in a larger sample.

Key points

Question: Is this brief group intervention feasible to address gaps in local service provision?

Findings: Statistical differences were found between attenders and non-attenders in addition to those completing the brief intervention. **Importance:** There may be utility in using health beliefs as a screening tool to identify those most at risk of non-attending; Increased knowledge of FND and improvements in health beliefs and quality of life were present 3 months after the intervention. **Next Steps:** To continue data collection in a larger sample and develop patient and clinician resources based on the content.

Keywords

Functional Neurological Disorder; brief intervention; psychological intervention; health beliefs; quality of life

Introduction

Functional Neurological Disorder (FND) refers to neurological symptoms that develop which have no known organic pathology, they are real and not imagined or feigned (Stone, Carson & Sharpe, 2005). These symptoms can present in various ways and those living with FND may experience multiple symptoms which can be purely functional or there may be a functional overlay to co-morbid organic health conditions. For example, limb weakness, abnormal movement, non-epileptic seizures, cognitive and/or sensory problems. The severity of symptoms found in FND is like other neurological conditions where symptoms can range from mild to severe with some patients being physically incapacitated as a result of the condition (Healthcare Improvement Scotland, [HIS] 2012).

However, diagnosis can be difficult with healthcare professionals trained to diagnose and treat disease rather than symptoms (Stone et al., 2005). As a result, if no disease is found there has been a suggestion historically, from healthcare professionals, that the symptom must not be real or the problem therefore must be psychological which is not the case (Stone et al., 2005). Messages to the brain from the body or from the body to the brain results in the symptoms the participants experience. Neurologists make the diagnosis of FND based on positive signs. For example, a person may present with limb weakness, reporting they have difficulty moving their leg when they try to carry out this movement, however when the person is asked to move their other leg the leg they have difficulty with reacts to the automatic reflex generated and responds as it should. This reflex reaction is Hoovers sign, an example of one of the positive signs Neurologists use in diagnosing FND. These positive signs indicate the neural connections between the brain and the muscle are intact demonstrating there is

not a structural problem within the nervous system. However framing symptoms as a result of biopsychosocial factors is preferred to a purely psychological aetiology (Stone et al., 2005).

It is estimated that one third of patients seen in Scottish neurology clinics have FND (Stone et al., 2005) making patients with FND one of the largest clinical groups accessing neurology services. The volume of patients presenting with FND presents a significant challenge to the healthcare services. Healthcare Improvement Scotland estimated the cost for the economy and healthcare service providers at £1.3 million for outpatient services, £6.01 million for inpatients and £4.01 million for primary care services (HIS, 2012). In addition, it is estimated that 27% of those with FND were not working due to their condition and are potentially more likely to be in receipt of state benefits creating significant financial demands on the state to fund and potential financial difficulties for those living with the condition (HIS, 2012).

Unlike other neurological conditions less is known about FND and typically there is no set treatment pathway for health boards to follow. This lack of treatment options to those with FND may start to change as clinical guidelines have begun to emerge. Stepped care is a recommended model, in a number of conditions such as common mental health disorders e.g. depression; generalised anxiety disorder (GAD); panic disorder; obsessive-compulsive disorder (OCD); post-traumatic stress disorder (PTSD) in addition to simple phobias (Kendrick & Pilling, 2012). Stepped care treatment allows patients to receive the level most effective for their difficulties at an appropriate level of resource, increasing the level of input when clinically required. A stepped care approach has been recommended as an appropriate model of treatment for FND (See figure 1; adapted from HIS, 2012).

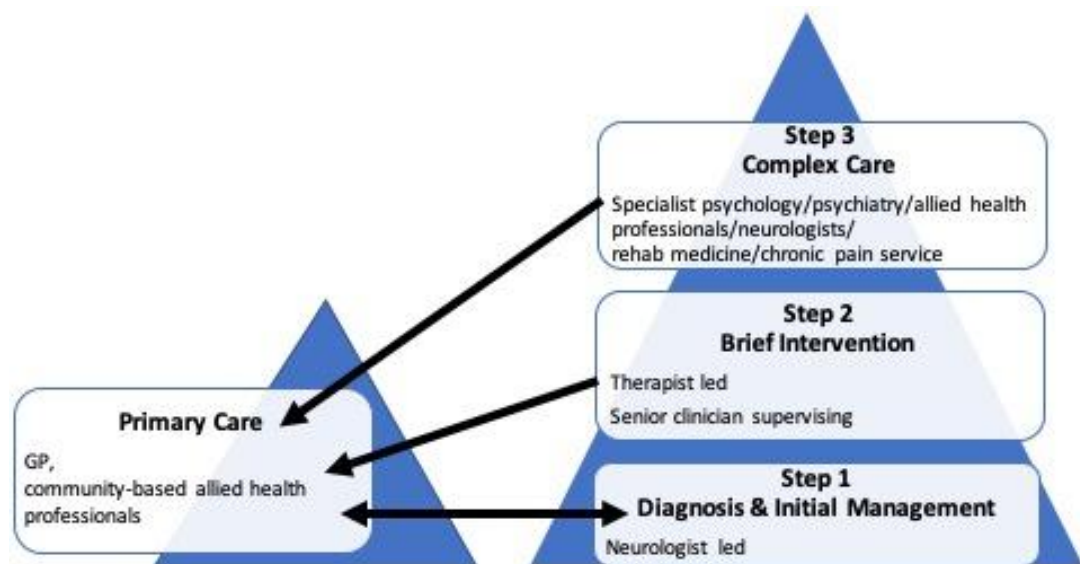


Figure 1 Stepped Care Model (adapted from HIS, 2012)

In the stepped care model proposed by HIS (2012) there are 3 broad steps for treatment which connect back into primary care community services; diagnosis and initial management; brief intervention and complex care. In step 1 the importance of a robust diagnosis of FND by the clinician was recommended and augmented with high quality patient information. HIS (2012) recognised that clinicians varied in their skills and recommended access to continuous personal development (CPD) learning for clinicians.

Step 2 aimed to meet the needs of patients with moderate physical disability and/or comorbid symptoms of mild/moderate anxiety/depression. People meeting these criteria should have access to brief intervention programme based on the principles of cognitive behavioural therapy (HIS, 2012). These services should be aiming to evaluate both the efficacy and cost effectiveness of these interventions (HIS, 2012). Patients requiring Step 3 were more likely to have severely levels of physical disability and/or complex mental health comorbidities (HIS, 2012). Step 3 could be accessed

directly if this was the level of input required or could be undertaken after steps 1 and 2 are completed (HIS, 2012). Allied health professionals at this level would be FND specialists assisting patients with managing their symptoms (HIS, 2012).

The recommendations made by HIS (2012) were augmented in a report from the BPS Division of Neuropsychology (2013) who recognised that although the stepped care model proposed in the earlier work was helpful, it lacked detail as to how patients would be matched to the level of care that best reflected their needs. The high personal costs to patients in terms of reduced physical function and quality of life due to clinical needs not being met effectively was also highlighted (BPS Division of Neuropsychology, 2013). The heterogeneous pattern of difficulties those with FND require knowledgeable and trained staff (BPS Division of Neuropsychology, 2013; HIS, 2012). A formulation-based approach was recommended, as was access to a centralised point of contact for both patients and clinicians who may be looking for appropriate information to assist patients with their diagnosis and access treatment processes which would be matched to the patients level of need (BPS Division of Neuropsychology, 2013).

A more recent addition to the guidelines for clinician's treating FND can be taken, indirectly, from new guidance for those working in non-specialist settings (National Institute for Health and Care Excellence [NICE], 2019). Although not a guideline specifically for FND, it is acknowledged and listed as a potential explanation for conditions such as reoccurring dizziness, reoccurring limb and facial weakness, numbness/tingling, difficulties with word finding and problems with memory (NICE, 2019). A further recommendation states that patients should be advised it is common in FND that symptoms can come and go and may fluctuate (NICE, 2019). The influence

of potential comorbid conditions on the symptoms of FND is also highlighted for example, anxiety and cognitive problems such as memory and word finding difficulties (NICE, 2019). It is recommended these difficulties are explored when assessing neurological conditions that are functional in nature (NICE, 2019).

Taken together these recent guidelines provide the beginnings of a framework for clinical implementation. There is a degree of overlap with their findings but a lack of clarity as to how these recommendations can be imbedded into clinical activity that can be managed within the limits of current resources. The importance the diagnosis process and patient information has been highlighted by guidelines as important, if this information is missing at diagnosis a stepped care model of care may be a suitable platform to provide this within a brief intervention (HIS, 2012). Overall there appears to be a lack of consensus and direction from clinical guidelines with regards providing effective treatments for those with FND after diagnosis. The interventions that are available are heterogenous and variables of interest vary widely with outcome measures for a study often being condition specific (BPS Division of Neuropsychology, 2013). For example with the primary outcome measure for interventions aimed at treating Non-Epileptic seizures focusing on the frequency of seizures (Barry, Wittenberg, Bullock, Michaels, Classen & Fisher, 2008; Baslet, Dworetzky, Perez, & Oser, 2015; Cope, Smith, King & Agrawal, 2017; Libbon, Gadbaw, Watson, Rothberg, Sillau et al., 2019; Prigatano, Stonnington & Fischer, 2002; Rusch, Morris, Allen, & Lathorp, 2001; Zaroff, Myers, Barr, Luciano & Devinsky, 2017). It may be more useful to explore the symptoms of FND using outcome measures that are more general in nature with a focus on areas highlighted in clinical guidelines such as NICE (2019).

Cognition in FND

Evidence about the importance of common comorbidities people with FND experience and ensuring these symptoms are considered when people present with neurological conditions that are potentially functional is starting to emerge (NICE, 2019). For example, many people frequently report co-morbid difficulties with cognition (Pennington, Newson, Hayre, & Coulthard, 2015). A recent review identified the main issues those with FND present with are poor attention, memory and language difficulties, namely problems with word finding (Teodoro, Edwards & Issacs, 2018). Despite the prevalence of difficulties and new recommendations to assess cognitive symptoms from clinical guidelines very few studies include a cognitive assessment battery in their protocol (Teodoro et al., 2018). This omission is surprising considering a correlation between impaired cognition and symptoms of depression have been found in this population previously (Teodoro et al., 2018). Cognitive difficulties have also been found to impact on quality of life for those with FND (Vechetova, Slovak, Kemlin, et al., 2018).

Mood in FND

Mental health difficulties are a frequent comorbidity in FND although it is common for those affected to contest referral to mental health services for treatment (Monzoni, Duncan, Grunewald & Reuber, 2011). However, evaluating symptoms of depression has been undertaken with many different types of outcome measures. For example studies have previously used the Hospital Anxiety and Depression Scale (HADS; Sharpe et al., 2011); The Depression Anxiety and Stress Scale (DASS-21; Barrett-Naylor, Gresswell & Dawson., 2018); Beck Depression Inventory (BDI; Barry et al., 2008; Baslet, et al., 2015) and Patient Health Questionnaire 9 (PHQ 9; Cope et

al., 2017; Libbon et al., 2019) which makes it difficult to generalise effects not only across research but also across care settings. Certain measures such as the HADS and the BDI are copyrighted for use which may exclude some services in the NHS having access to funds to pay for it in routine clinical practice.

Health Beliefs in FND

In addition to evidence about mood and cognition other common psychological constructs have been identified that can impact on the patient's recovery from FND. The first of these are the beliefs patients attach to their symptoms to make sense of their experience. The valence of health beliefs in FND has been identified as an important predictor of patient progress 12 months later (Sharpe, Stone, Hibberd, Warlow, Duncan., et al., 2010). Furthermore, those with negative health beliefs have been found to be less reassured when test results are normal (Donkin, Ellis, Powell, Broadbent, Gamble & Petrie, 2005) which might hold consequences for healthcare care services. For example, if patients can not readily accept that there is nothing untoward found in the investigations, they may seek additional appointments with clinicians for reassurance or further investigations. Negative health beliefs are common in those experiencing other illnesses with a functional basis such as chronic fatigue (Knoop, Prins, Moss-Morris & Bleeijenbergh, 2010), fibromyalgia (Glattacker, Opiz & Jackel, 2010) and irritable bowel syndrome (Rutter & Rutter, 2002).

Health Related Quality of Life in FND

Another psychological construct explored frequently in FND research is quality of life. Quality of life is a difficult domain to define but is generally considered to be a multifaceted (Siegrist & Junge, 1989). Health-related quality of life (HRQoL) refers not only to an individual's overall ability to function but also the subjective perception they

have regarding the quality of their experience within the physical, mental and social domains of their life (Hays & Morales, 2001). Function in HRQoL refers to the ability to perform activities of daily living such as personal care (e.g. washing and dressing), work (e.g. employment or household chores) and social function (interactions with friends and family members) (Hays & Morales, 2001).

Those with FND often report a poor quality of life due to living with restricted physical function (BPS Division of Neuropsychology, 2013). A high percentage of people find they must stop work due to the debilitating nature of their condition on their function resulting in many people with FND relying on state benefits (BPS Division of Neuropsychology, 2013). Other symptoms found to negatively on HRQoL for those with FND include fatigue (Gelauff, Kingma, Kalkman et al., 2018; Vechetova, Slovak, Kemlink, Hanzlikova, Dusek et al., 2018) depression/anxiety, insomnia, pain and difficulties with cognition (Vechetova et al., 2018).

There are many ways to measure HRQoL one of which is the utility measurement approach. In this method a ‘utility’ is a value that can be derived from a current health state where ‘1’ is the best health imaginable and ‘0’ is equal to death (Sassi, 2006). Life quantity can be measured in how many years there are between birth and death and the quality of life is ascertained at different timepoints between the endpoints of birth and death (Torrance, 1987). An important variable in ascertaining the balance between life expectancy, quality of life and treatment related costs is a unit of measurement called a quality-adjusted life-year (QALY). These units can be considered as being indicative of the “quality of a lifetime” (Torrance, 1987).

QALY scores can be extracted from quality of life measures such as the Short Form 36 (Ware & Sherbourne, 1992), although it is felt that these measures may

underestimate QALY gains, therefore the recommended tool for conducting a cost utility analysis is the EuroQoL-5D (EQ-5D; Yang, Devlin & Luo, 2019). The EQ-5D is also recommended by the National Institute for Health and Clinical Excellence (NICE) for calculating quality adjusted life years (QALYs) (NICE, 2008). The QALY is an index of the effectiveness of an intervention calculated by measuring the impact on life expectancy and quality of life of the patient compared to their current experience. The QALY summarises the patients' response on a standardised outcome measure that represents quality of life in several domains. The length of time a patient has experienced the condition is used in the pre-treatment calculation and post intervention it is the length of time benefits from treatment are expected for that is used. For example, if a patient manages a condition for 5 years with no intervention and their current ratings result in a utility value of 0.8 the patient will have 4 QALYs ($5 \text{ years} \times \text{utility value of } 0.8 = 4 \text{ QALYs}$). If the patient, then accesses a new treatment and their ratings on the measure result in an increased utility value of 0.95 the patient will then have 4.75 QALYs ($5 \text{ years} \times \text{utility value of } 0.95 = 4.75 \text{ QALYs}$). As there has been an increase of 0.75 in the QALY post intervention, the intervention will be deemed to have a value of 0.75 QALYs over the no treatment option.

Summary

Quality of life experienced by those with FND may be negatively affected by the lack of treatment options after diagnosis. There are currently no FND specific treatments available within NHS Grampian after diagnosis. This research project aims to evaluate delivering a brief two session intervention to ascertain if this would be a feasible method by which to address a gap in local services and offer an effective

treatment option for people with FND within routine clinical practice in NHS Grampian.

The recommendations in the clinical guidelines produced to date for FND (BPS Division of Neuropsychology, 2013; HIS, 2012; NICE, 2019) have informed the psychological variables selected for this study. These include cognitive function, mood, health beliefs and quality of life. In addition to being recommended as domains being particularly relevant to FND, cognitive function, mood and health beliefs have all been found to impact negatively on quality of life for those with FND in previous research (Cope et al., 2017; Sharpe et al., 2010; 2011; Vechetova et al., 2018). The healthcare utilisation and a cost utility analysis informed the feasibility variables. Finally, this study will explore the expectations, knowledge and experience of those taking part in the intervention

This research will allow a unique opportunity to explore potential differences between participants who attended and did not attend the intervention in addition to those who completed the programme. The results from the outcome measures used in this feasibility project will be a chance to review the outcomes administered in the FND population which holds potential to inform outcome measures used as part of service development and possibly inform future research programmes with NHS Grampian.

Study Aims

This research aims to collect data over 4 timepoints. Baseline (T1), Pre-Group (T2), Post-Group (T3) and 3 months after the group, Follow-Up (T4). The research questions informing the current study are as follows:

1. Did those attending the group differ significantly from non-attenders in their demographic, psychological or healthcare utilisation profile?
2. What changes occurred in the standardised outcome measures indexing psychological constructs over the four timepoints, for those completing the group intervention?
3. Did healthcare utilisation and healthcare costs vary for those attending the group intervention between T1 and T4?
4. Did the experiential aspect of the group meet the expectations of those participating in the group intervention and were significant improvements reported in subjective levels of knowledge about FND post intervention?

Methods

Participants

Potential participants with FND were identified by the Consultant Clinical Neuropsychologist (FS) supervising the project from the departmental database at Aberdeen Royal Infirmary. This database was pre-existing and was not part of this research, it is maintained by the Department of Clinical Neuropsychology. It contained details of around 200 patients who had received a diagnosis of FND or were being seen and had a pre-existing diagnosis between November 2018 and April 2019 within the neurology service at NHS Grampian. The data for the database was input by an assistant psychologist from the Department of Clinical Neuropsychology from referral sheets received from the consultant neurologists. Information included demographics such as age, gender, postcode; source of the referral to neurology; type of FND, whether it was pure FND or a functional overlay to another neurological condition; patient comorbidities; the other specialties the patient has accessed; number of emergency consultation and admissions; number and type of radiology investigations; number of medications.

Inclusion Criteria

To ensure the participants were as close to patients that would be seen in routine clinical practice as possible, there was only one criterion for inclusion which was potential participants had to be aged between 18-64 years with a diagnosis of FND from a consultant neurologist.

Exclusion Criteria

Exclusion criteria were as follows:

1. Anyone the clinical neuropsychologist/neurologist felt, as a result of their clinical judgement, lacks capacity to consent.
2. Anyone that was unable to understand the information sheet due to English not being their first language. This may be problematic as the information within the study relies on an understanding of written and spoken English. If knowledge of the English language is limited there would be a difficulty understanding the information being delivered.
3. Severe sensory impairment.
4. Anyone experiencing active severe psychiatric symptoms.
5. A dependency on alcohol or recreational drugs.
6. Learning disability as classified by ICD 10.
7. Those unable to travel independently to attend group sessions due to the severity of their symptoms.

Design

This research was a feasibility study to develop future therapeutic options patients may have access to as part of a new care and treatment pathway within NHS Grampian. Participants acted as their own control as baseline measures (T1) were collected depending when participants were seen, the T2 pre-group measures were completed at the start of the group intervention. At the beginning of the appointment the participant information sheet was reviewed with the participant and an opportunity was given for any questions the participant may have had. Confidentiality and time

commitment of the study were discussed prior to informed consent being taken (see Appendix 2) the baseline outcome measures were then completed.

Recruitment Procedure

Prior to contacting patients from the database, the consultant clinical neuropsychologist reviewed the database for names for patients diagnosed with FND by consultant neurologists. The consultant neurologist who originally referred the patient for inclusion on the database reviewed those patients as potential participants to ensure they were eligible to take part. Invitation letters were prepared by the researcher which were signed by the consultant neurologist that had overseen the participants care and sent along with a participant information sheet, a reply sheet and a pre-paid envelope (See Appendix 3). If an individual was interested in the research, they notified this by returning a reply slip to the Department of Clinical Neuropsychology. Potential participants were allocated an individual reference number to allow for information that was being returned could not be explicitly identified.

On receipt of the reply sheet the researcher (PI) used the identification number to find the potential participants name and contacted them using telephone or email depending on the preference stated in the reply. This initial contact gave the participant the opportunity to discuss the research and ask questions they may have had. If the person wished to take part in the research study a baseline appointment was arranged. In total, 29 baseline (T1) appointments were completed by the researcher (PI). At baseline (T1) participants were given the opportunity to raise any questions they may have had regarding the study. Once the participant had no further questions or queries informed consent was taken. Baseline questionnaires covering the psychological and

feasibility outcomes were then completed, more details about these are given later in this section.

The Group Intervention

Group Allocation

As a result of availability of an appropriate room and the field supervisor (FS) there were 4 groups available. Group A and B were run on Tuesday morning and afternoon respectively, Group C was Wednesday morning with Group D on Wednesday afternoon. Participants self-selected the group day and time that was most convenient for them to attend. After baseline measures were completed 29 participants were distributed as follows over the four available group slots; [Group A (n=7); Group B (n=8); Group C (n=7); Group D (n=7)]. Attrition rates were high [45% (n=13)] between T1 and T2 resulting in a reduction in the number of people within the groups who attended the first session a further 19% (n=3) of those attending session 1 not attending session 2 (T3) two weeks later a further two individuals were lost to follow up resulting in 11 participants completing the intervention [Group A (n=1); Group B (n=3); Group C (n=4); Group D (n=3)]. There were no differences in the format or content of the groups all participants received the same intervention content, delivered by the same facilitators in the same location.

Group Design

This brief intervention for FND in this feasibility study was comprised of two group sessions. Sessions were split into two to minimise participant fatigue and allow an opportunity to reflect and consolidate the information presented. The sessions lasted for 90 minutes and were spaced a fortnight apart. The aims of the group were based on a biopsychosocial model. A formulation approach was followed putting symptoms in

the context of predisposing, precipitating and perpetuating factor. The concept of how biological and psychological factors potentially combined in FND was also introduced to explain why symptoms potentially occurred and why symptoms may fluctuate.

The aims were to support participants to:

1. Understand more about Functional Neurological Disorder.
2. Develop knowledge about how physical and psychological processes interact in the body.
3. Build knowledge of effective coping strategies.
4. Develop awareness about things that can make symptoms worse.
5. Gain awareness of resources and support available.
6. Give participants opportunity to meet with others who have been through similar experiences.

Group Resource Development

Several resources utilised in the group intervention were developed specifically for the study by the researcher (PI) under the supervision of the consultant clinical neuropsychologist (FS) supervising the project within NHS Grampian.

Group Content

A formulation-based approach was used to frame FND from a biopsychosocial perspective. PowerPoint slides for both sessions were developed and due to limited time, these were also printed to serve as participant handouts. Flipcharts were used to facilitate discussion and capture information on common symptoms, potential triggers, what made symptoms worse and what helped.

The first session focused on what FND is and gave a basic overview of the central and peripheral nervous systems using a diagram presented on PowerPoint. Participants were given opportunity to discuss their thoughts around the complexity of the brain/body connection. Common symptoms were explored, and the group was invited to discuss this. Potential triggers for FND were then discussed giving the group chat opportunity to share their individual experiences.

The second session content centred on living with FND included information regarding fatigue management, sleep, cognition, anxiety and depression and possibility of recovery. These topics are transdiagnostic with other neurological conditions therefore the content of the FND group was based on similar groups run within the department of Clinical Neuropsychology in NHS Grampian for Acquired Brain Injury and Multiple Sclerosis.

Consultant Videos

Bespoke videos for the study were developed by the researcher (PI) under the supervision of the field supervisor (FS) to augment the group content. Two consultant neurologists working in NHS Grampian were invited to be recorded answering questions about FND. Those attending baseline appointments for the group intervention also guided content from the questions and conversation held with the researcher (PI) during their session. The video was shown in sections, the neurologists recorded content on what FND is, the diagnostic process, the likelihood of misdiagnosis, advice on living with FND and the chance of recovery.

Filming was completed by the audio-visual department of the University of Aberdeen and took place within neurology department in NHS Grampian. After the

research was completed the videos were utilised as a source of information for patients receiving a diagnosis. The videos can also be accessed from a QR code in the health boards information leaflet and are also hosted on this website (see <http://www.nhsgrampian.org/neurology/FND.html> to access the videos and see a copy of the leaflet).

Facilitator Guide

A facilitator guide for the group was also developed. The guide had 24 pages consisting of information about the group aims and background literature the intervention was based on. The slides for both sessions were included with prompts regarding the key messages for each slide in order to assist consistency should the intervention be delivered in the future.

Participant Feedback Questionnaires

Two bespoke questionnaires were developed See Appendices 4 and 5 The first measured participants subjective changes in knowledge levels pre and post group. The second was more general and captured participants subjective experience of the group both in terms of content and delivery.

Group Outcome Measures

1. Demographic Information

This included age, post-code, gender, marital status, years of education, symptom duration and age when symptoms began (see Appendix 6). The Consultant Clinical Neuropsychologist (FS) provided categories to group types of symptoms based on the participants diagnosis, these were sensory, cognitive, motor, altered awareness and non-specific clinical presentations (see Appendix 7 for more information) . The Scottish Index of Multiple Deprivation code (SIMD) was derived from post code to

provide an index of socioeconomic status. Demographic information was collected at baseline (T1) only.

2. Cognitive Function: The Epitrak

The Epitrak (Lutz & Helmstaedter, 2005) was utilised as a brief measure of cognitive function, it contains six subtests measuring response inhibition, visuo-motor speed, mental flexibility, visuo-motor planning, verbal fluency, and working memory. Administration time was around 15 minutes. The total score is age corrected against a normative sample with higher scores indicative of a better performance and contains an indicator of clinical change as part of the scoring; the maximum score is 49 points. Scores between 29–31 points indicate mild impairment, Scores below 28 points are indicative of moderate impairment of cognitive function in these domains. This measure has been found to have good reliability and criterion validity (Lutz & Helmstaedter, 2005). The EPI-Trak was administered at baseline (T1) and follow up (T4).

3. Psychological Measures

Standardised questionnaires indexing symptoms of depression (PHQ-9; Kroenke, Spitzer & Williams, 2001); anxiety (GAD-7; Spitzer, Kroenke & Williams, 2006); health related quality of life (SF-36; Ware & Sherbourne, 1992), and health beliefs (IPQ-B; Broadbent, Petrie, Main & Weinman, 2006) were completed at 4 timepoints. These were prior to the intervention (baseline, T1) at the start of the 1st group session (pre-group, T2). The third time point was immediately after the intervention (post group, T3), the fourth and final timepoint was 3 months after the group (follow up T4).

Mood: Depression PHQ-9

Symptoms of depression were assessed using the Patient Health Questionnaire 9 (PHQ-9; Kroenke et al., 2001). Nine items assess experience of depressive symptoms over the previous 14 days, ranging from not at all to nearly every day, total score ranges from 0-27 with higher scores indicating more severe depressive symptoms. Scoring 5, 10, 15, and 20 categorises depressive symptoms into mild, moderate, moderately severe and severe respectively. This measure has been shown previously to have good validity and reliability (Kroenke et al, 2001).

Mood: Anxiety GAD-7

Anxiety symptoms were measured using Generalized Anxiety Disorder (GAD-7; Spitzer et al., 2006). This measure has 7 items with 4 options measuring levels of anxiety over the previous 14 days ranging from not at all to nearly every day with scores ranging from 0-12 with cut-offs of 5, 10, and 15 representing cut-off scores for mild, moderate, and severe anxiety, respectively. The measure has good reliability and validity (Spitzer et al., 2006).

Both the PHQ-9 and GAD-7 have been used to assess levels of depression and anxiety in participants with functional neurological disorders taking part in group psychological interventions previously (Cope et al., 2017; Libbon, et al., 2019). Both measures are self-report based on the participant's perception of symptoms experienced over the previous 14 days, rating whether they experience difficulties nearly every day to not at all. Both measures are recommended by The National Institute for Health and Care Excellence (NICE; Clark, 2011) and were completed at T1, T2, T3 and T4.

Health Related Quality of Life SF-36

The SF-36 (Ware & Sherbourne, 1992) consists of 36 items with between 2 and 6 responses to select from, each item has a minimum score of 0 and a maximum score of 100. 35 of the individual items are grouped together to give 8 subscales: physical function (10 items), physical health limitations (4 items), emotional health limitations (3 items), energy/fatigue (4 items), emotional wellbeing (5 items), social function (2 items), pain (2 items) and general health (5 items). Subscales are scored by adding the scores of all items within the subscale together and dividing by the number of items in the subscale to get a mean score for each domain with higher scores indicating a higher quality of life (Ware & Sherbourne, 1992). The SF36 has previously been found to show good criterion validity and high internal reliability (Jenkinson, Wright and Coulter, 1994), it has also been used in populations with FND previously (see Pick et al., 2020 for a review). The SF-36 was completed in the current study at T1, T2, T3 and T4.

Health Beliefs: IPQ-B

The Brief Illness Perception Questionnaire (IPQ-B) has been found to show good validity and reliability (Broadbent et al., 2006). The IPQ-B has been utilised in assessing the health beliefs of people with functional neurological disorders in previous group psychological interventions (Cope et al., 2017; Libbon et al., 2019). This measure of health beliefs has nine items and is based on components of the Self-Regulation Model (SRM) (Leventhal et al., 1980; 1984) and assessed both cognitive and emotional representations patients had about their FND. The IPQ-B contains 5 items on cognitive factors included in illness perception including: consequences (item 1), timeline (item 2), personal control (item 3), treatment control (item 4), and identity

(item 5). Illness concern (item 6) is the perception of both cognitive and emotional beliefs. The scale contains 2 emotional items; coherence (item 7) and emotional representation (item 8). Participants rate each item on a 0–10 scale. The last item in the brief IPQ allows participants to identify factors they believe are causal in the development of their FND symptoms for categorical analysis. Levels of threat perceived by the participant can be calculated by reversing the scores from item numbers 3, 4 and 7 and adding to other items. Higher score indicates higher perceived threat with a minimum threat level of 0 and a maximum level of 80. The IPQ-B was administered at T1, T2, T3 and T4 of the study.

4. Healthcare Utilisation and Costs Measurement –

These outcomes were included to build an understanding of the frequency patients with FND accessed health care services allowing calculation of the associated costs. Health Care Utilisation was collated using the Client Service Receipt Inventory; (Chisholm, Knapp, Knudsen, Amaddeo, Gaite, et al., 2000)

Healthcare Utilisation: The Customer Services Receipt Inventory

The Customer Services Receipt Inventory (CSRI) was utilised as a measure of healthcare use over the previous 3 months. The CSRI has been found previously to offer a standardised and adaptable measure that can be individualised for the population being studied (Chisholm et al., 2000). In the current study the CSRI included healthcare utilisation in three health sectors; Community care, Elective care and Non-Elective Care. The community care category recorded the number of contacts participants had with their GP and/or practice nurses; the number of repeat prescriptions was also collated. Elective care included outpatient appointments with specialties that were most likely to be linked to someone with FND, this section also

included pre-arranged hospital contacts as an overnight case, an outpatient and/or a day patient, finally non-elective care provision use was measured. Further information regarding medication, formal support from the NHS/Social Work and informal support from friends and family as well as employment, sick leave, earnings and state benefits accessed. The CSRI has been used as a measure of healthcare utilisation for people with FND previously (Goldstein, Chalder, Chigwedere, Khondoker, Moriarty, et al., 2010).

HRQoL Cost Utility Analysis: EQ-5D

The EQ-5D is a 2-part self-report questionnaire with 2 components; a descriptive system to calculate health state and a visual analogue scale (VAS) (Herdman, Gudex, & Lloyd et al.. 2011). The health state component has five categories: mobility, self-care, usual activities, pain/discomfort and anxiety/depression with 5 options to choose from: no problems, slight problems, moderate problems, severe problems and extreme problems, these options are scored from 1-5 respectively with higher numbers reflective of greater impairment. Health state responses are indicated by ticking the most relevant of the 5 options, the score from each option is compiled into a 5-digit number representing the health state which is converted into a utility value (quality of life). The utility value can fall between 0 (Dead) and 1 (Perfect Health) and can be calculated for people in the UK using a calculator available from the website of the EQ-5D developers

(see <https://euroqol.org/eq-5d-instruments/eq-5d-5l-about/valuation-standard-value-sets/crosswalk-index-value-calculator/>). The quality adjusted life years (QALY) is derived from the utility value by multiplying it with the number of years a person has lived with a condition. The EQ-5D has been used to evaluate cost utility outcomes in

patients with functional disorders previously in a physiotherapy treatment programme (Nielsen, Ricciardi, Demartini, Hunter, Joyce, & Edwards., 2015) and is the recommended by NICE as the instrument of choice for QALY generation (NICE, 2008).

The VAS in the EQ-5D is labelled ‘The best health you can imagine’ at the top of the scale and ‘The worst health you can imagine’ at the bottom. Best health has a maximum score of 100 with worst health minimum scoring is 0. A mark is placed on the scale and the corresponding number written in the box alongside. The EQ-5D was administered at baseline (T1) and follow up (T4).

5. Experiential Measures – At end of the group and at follow up people were encouraged to feedback their experience of taking part in the group intervention by completing bespoke questionnaires to capture this feedback.

Participant Knowledge

Those attending their first group session were invited to self-report the following using a five-point Likert scale (see Appendix 4)

1. their current levels of knowledge levels what they thought FND is,
2. the process of how FND was diagnosed;
3. how much they knew about how physical & psychological processes interact in the body
4. what they knew about effective coping strategies;
5. Knowledge levels on what made symptoms get worse; what they knew about resources that were available.

Participants rated their knowledge levels on a 5-point Likert scale with options ranging from 'I know nothing' to 'I know a lot'. Participants chose the box they best felt reflected their current knowledge level for each area and indicated how helpful they thought meeting others with FND would be. This exercise was repeated after the delivery of the group.

Participant Experience

At the end of the second session, participants were also given the opportunity to complete anonymous generic feedback on the workshop (See Appendix 5). The items covered how acceptable the group intervention was to those taking part in terms of the number of sessions, their duration, what was included and length of time between the sessions. Participants were also asked to rate their thoughts on the quality of the location and group facilitation. Additional questions gave opportunity for participants to feedback their thoughts on what they liked or would change about the content. Finally, participants were asked if they would recommend the group to a friend with FND if it were available. The study matrix provides a summary of all the measures and the time points these were administered during the study (see Table 1). All outcome measures were anonymised using a sequential participant ID number.

Table 1 Study matrix table of measures and administration timepoint

	Stage 1 Study Preparation	Stage 2 Pre-Group Baseline Phase T1	Stage 3 Group Phase Pre- Group T2	Post- Group T3	Stage 4 Longitudinal Evaluation Phase T4
University sponsorship, ethics (IRAS and University)					
NHSG R&D Approval	*				
Develop Group materials	*				
Informed consent		*			
DEMOGRAPHICS		*			
COGNITIVE FUNCTION					
Epi-Trak		*			*
PSYCHOLOGICAL MEASURES					
Mood: Anxiety		*	*	*	*
Generalised Anxiety Disorder 7 (GAD 7)					
Mood: Depression		*	*	*	*
Patient Health Questionnaire 9 (PHQ-9)					
Quality of Life		*	*	*	*
Short Form health Survey (SF-36)					
Health Beliefs					
Brief Illness Perception Questionnaire (IPQ-B)		*	*	*	*
FEASABILITY MEASURES					
Healthcare Utilisation		*			*
Client Services Receipt Inventory (CSRI)					
Cost Utility Analysis		*			*
EuroQoL-5D (EQ5D)					
EXPERIENTIAL MEASURES					
Patient Knowledge			*	*	
Patient Experience				*	

Ethical Approval

A favourable ethical opinion was granted by NHS North of Scotland Research Ethics Service (REC Reference 18/NS/0137; Approved 17 January 2019; See Appendix 10). The research was also registered with the NHSG Research & Development Office receiving management permission to proceed locally with the research (R&D Reference 2018PC011; Approved 19 January 2019; See Appendix 11).

Statistical Analysis

Data Screening

Statistical analysis was undertaken using IBM Statistical Package for Social Sciences version 24. Data was explored in line with the recommendations given in Field (2005) to establish the distribution of the data in order to ensure it met criteria for using parametric tests in analysis. All variables in the standard measures met assumptions for parametric analysis except the SF36 subscale for physical limitations for Pre-group (T2) only which was positively skewed ($Z_{\text{skewness}} > 2.58$; Field, 2005). This variable was transformed using the square root transformation in SPSS 24 to restore normality ($Z_{\text{skewness}} = 2.00$; Field 2005) prior to analysis.

In order to establish if duration of symptoms correlated with psychological outcomes Pearson's correlations were undertaken (see Appendix 1). There were no significant correlations detected between the potential covariant and cognitive function (Epi-Trak), depression symptoms (PHQ-9); symptoms of anxiety (GAD-7); subscales of measures tapping health beliefs (IPQ-Brief) and HRQoL (SF-36) as a result symptom duration was not considered further in the analyses.

Boxplots were used to visually inspect the data for outliers. Z scores for each of the dependent variables for the standard measures were calculated using SPSS in order to screen for potential outliers. The critical value to establish if a participant could be considered an outlier was a z-score that was either less than -3.29 or greater than +3.29. This critical value was adopted as it would be unlikely data that were normally distributed would fall out with this limit (Field, 2005). Identifying the cause of outliers is not a straightforward process in clinical research, and there has been debate on data sets ought to be managed. Consequently, researchers have been encouraged to apply their own judgement as to whether outliers are removed or included in data (Osborne and Overbay, 2004).

Data Analysis: Attenders and Non-Attenders

If a participant completed base line measures and attended the first group session they were categorised as an attender. Those completing baseline measures but not attending the first session were categorised as a non-attender. Analysis for study aim 1 which explored differences between attenders (n=16) and non-attenders (n=13) on continuous demographic variables were conducted using independent samples t-tests. As sample sizes were small, categorical variables in the demographic information were analysed using a Fishers Exact test. Independent samples t-tests were also used to compare attenders and non-attenders on psychological outcome measures.

Due to some healthcare providers being accessed only once or not accessed at all, data from the Client Services Receipt Inventory were collapsed across provider representing healthcare utilisation within community, elective and non-elective care providers. The new variables provide a summary of the total number of contacts for both attenders and non-attenders for each of these 3 health sectors and independent t-

tests were used to explore if contact levels differed between attenders from non-attenders.

Experiential measures were not completed by non-attenders therefore it was not possible to explore differences regarding experience of the group.

Data Analysis: Completers

Due to the brief nature of the intervention, a completer was classified as participant who completed measures at baseline (T1), pre-group (T2) and Post-Group (T3) and follow up (T4) at 3 months. Due the small sample size and the number of participants within each group [Group A (n=1); Group B (n=3); Group C (n=4); Group D (n=3)] data were collapsed across groups (n=11) for all analyses.

Data for study aim 2 (psychological measures) were analysed using a repeated measures ANOVA, Bonferroni adjustments were used for pairwise comparison. Paired sample t-tests were used to analysis changes for study aim 3 (healthcare utilisation) and study aim 4 (patient experience) outcomes.

A reliability analysis was carried out on the participant knowledge questionnaire created specifically for the FND brief intervention. A Cronbach's alpha (α) over .7 represents a high internal consistency. The six items assessing knowledge levels showed acceptable levels of reliability when administered pre-group ($\alpha = .76$) and post group ($\alpha = .81$).

Results

Recruitment

Recruitment began in February 2019 (see Figure 1 for flow of participants). 187 patients were identified by a consultant clinical neuropsychologist using the FND Database managed by the Department of Clinical Neuropsychology based in NHS Grampian. The Consultant neurologists aligned to the patient was contacted to review if the patient met study criteria prior to being invited to take part. As a result of the neurologists' review 35 patients were felt not to meet study criteria an additional 53 identified patients were not progressed within the study due to not receiving confirmation from their neurologist that they were eligible to invite. In total 98 letters of invitation were sent out to eligible patients in April 2019

There were 55 reply sheets received between April and August 2019. 77% of respondents (n=42) wished to participate in the research, 69% (n=29) completed baseline measures. These baseline appointments lasted approximately 60 minutes and were undertaken between June until September 2019 one morning a week within NHS Grampian. 19% (n=8) did not attend the baseline appointment, 5% (n=2) did not respond to contact following up their reply sheet and 7% (n=3) were not eligible. Of the participants who were ineligible two were hospitalised for psychiatric conditions during the recruitment window and one person revealed they were receiving treatment for an active substance misuse issue at the baseline appointment.

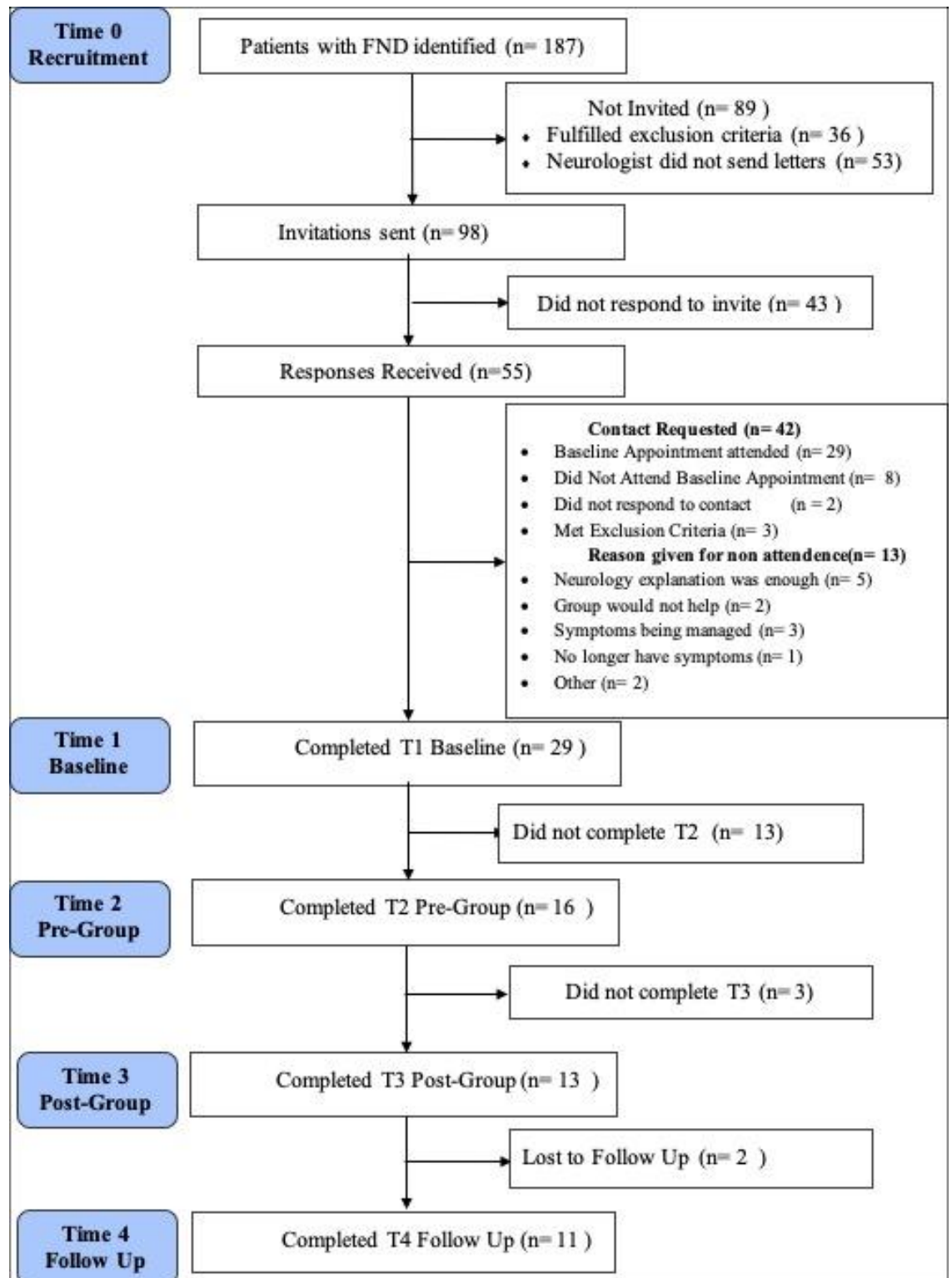


Figure 1 Study Participant Flow Diagram

Not all of those completing baseline measures (T1) attended the group intervention. The attrition rate between completing baseline and attending the first group session (T2) was 45% (n=13). with a further 19% (n=3) of those attending session 1 not attending session 2 (T3) two weeks later. 85% (n=11) of participants completing the group attended the follow up session in February 2020, 3 months post group (T4). The mean timescale from baseline to session 1 was 91.19 days (SD 31.38 days). No reasons were given for not attending by those leaving the research.

Participants

A total of 29 people completed baseline measures with 13 of those not attending the first group session. There were two different analyses conducted, the first included all participants who attended the baseline appointed and completed T1 measures. This analysis compared those participants who attended the first group session to those who did not. The second analysis included only those participants that completed T1, T2, T3 and T4 measures there by completing the group intervention and returning 3-month post group follow up.

Study Aim 1

The first aim of the current study was to explore if there were significant differences between attenders and non-attenders in demographic, psychological and healthcare utilisation profiles.

Demographic Information

The demographic information of all participants completing baseline (T1) measures can be seen in Table 2. There was no significant difference between attenders and non-attenders for any of the demographic variables.

Table 2 Demographic profile analysis for Attenders and Non-Attenders

	All Participants			Attenders			Non-Attenders			Independent Samples t-test (df=27)	
	M	SD	Min-Max	M	SD	Min-Max	M	SD	Min-Max	t	p
Age in years	46.34	(12.30)	21 - 77	47.88	13.750	25 - 77	44.46	10.47	21-58	-.74	.47
Age symptoms began	39.73	(14.76)	07 - 75	41.29	17.69	7 - 75	37.23	10.72	20-54	-.63	.53
Symptom duration (months)	86.31	(123.62)	12 – 492	83.81	150.93	12 - 492	89.00	87.73	12-240	.11	.91
Education in years	13.41	(2.11)	11 – 18	13.62	2.21	11 - 18	13.15	2.04	11-18	-.59	.56

Study Aim 1 Demographic profiles Attenders and Non-Attenders

Variables in Table 3 are represented by frequency data for all participants as well as attenders and non-attenders. The postcode provided by the participant allowed calculation of local area deprivation using the Scottish Index of Multiple Deprivation (SIMD) database to provide a measure of socioeconomic status. SIMD quintiles were used where SIMD quintile 1 has most deprivation and SIMD quintile 5 has the least deprivation.

Fishers Exact test was used for statistical analysis of these frequency variables as a result of the expected counts being less than 5 in the statistical comparisons of the frequencies. No statistical differences were found between those attending or not attending the group intervention on any of the analyses suggesting these variables were not a factor in determining who attended the intervention.

Table 3 Demographic frequency data for gender, marital status, SIMD Quintile and Geographical Locality, and employment status for all participants, group attenders and non-attenders.

	All Participants (n=29)	Attenders (n=16)	Non-Attenders (n=13)	Fishers Exact test P (two-tailed)¹
Gender (%)	20 Female (68.9%) 9 Male (31.1%)	10 Female (62.35%) 6 Male (37.5%)	10 Female (76.9%) 3 Male (23.10%)	0.45
Marital Status (%)				0.92
Married	17 (58.60%)	10 (62.50%)	7 (53.8%)	
Single	8 (27.60%)	4 (25.00%)	4 (30.8%)	
Widowed	3 (10.30%)	2 (12.00%)	1 (7.7%)	
Divorced/Separated	1 (3.40%)	0	1 (7.7%)	
SIMD (%)				0.87
1=Most Deprived 5 = Least Deprived				
SIMD Area 1	2 (6.90%)	0	2 (15.40%)	
SIMD Area 2	6 (20.70%)	4 (25.00%)	2 (15.40%)	
SIMD Area 3	9 (31.00%)	4 (25.00%)	5 (38.50%)	
SIMD Area 4	6 (20.70%)	3 (18.80%)	3 (23.10%)	
SIMD Area 5	6 (20.70%)	5 (31.20%)	1 (7.70%)	
Locality (%)				0.57
Aberdeen City	12 (41.40%)	5 (31.25%)	7 (53.80%)	
Aberdeenshire	9 (31.38%)	8 (50.00%)	1 (7.70%)	
Moray	8 (27.60%)	3 (18.75%)	5 (38.50%)	
Employment Status (%)				0.46
Full Time Work	6 (20.70%)	2 (12.50%)	4 (30.80%)	
Part-Time Work	6 (20.70%)	2 (12.50%)	4 (30.80%)	
Retired	3 (10.30%)	2 (12.50%)	1 (7.70%)	
Not Working (Health)	10 (34.50%)	7 (43.80%)	3 (23.10%)	

SIMD = Scottish Index of Multiple Deprivation

FND results in symptoms that are like disorders that have a structural or neurodegenerative cause, but the mechanism for people with FND is different. All participants had a diagnosis of FND from a consultant neurologist. For the purposes of this analysis the categories formulated by the Consultant Clinical Neuropsychologist were used to assess primary presenting FND symptom of participants (see Table 4).

Table 4 Frequency data for FND Presentation symptom for all participants

Main Presentation (%)	FND All Participants (n=29)	Attenders (n=16)	Non-Attenders (n=13)	Fishers Exact test P (two-tailed)¹
				0.22
Altered Awareness	5 (17.20%)	4 (25%)	1 (7.70%)	
Motor	7 (24.10%)	4 (25%)	3 (23.05%)	
Non-Specific	3 (10.40%)	0	3 (23.05%)	
Sensory	14 (48.30%)	8 (50%)	6 (46.20%)	

¹ Attenders & Non attenders

Comparisons between the type of FND symptom experienced by patients attending and not attending the group were conducted and no significant differences were detected using Fishers exact test ($p=0.22$) which can be interpreted as the main type of FND symptom the participant presented with did not determine whether they attended or did not attend the group intervention.

Psychological Measures: Attenders and Non-Attenders

Psychological outcome measures collected at T1 were analysed to determine if there were significant differences between attenders and non-attenders (see Table 5).

Cognitive Function: Epittrak

There were no significant differences between attenders and non-attenders found in the cognitive domain [$t(26) = -.81$; $p = 0.42$]; However, using Epittrak cut off scores to interpret the results showed participants would be categorised differently. Attenders ($M=32.07$) scored in the average range for overall performance whereas non-attenders fell into the mildly impaired category ($M=30.08$).

Mood: Depression PHQ-9 & Anxiety GAD-7

There were no significant differences found for depression, $t(27) = -.93$; $p=0.36$; or anxiety $t(27) = -.63$; $p=0.54$ between attenders and non-attenders. Interpreting the PHQ-9 scores in line with the clinical cut off scores showed both attenders ($M=14.50$) and non-attenders ($M= 12.08$) would be categorised as having moderately severe levels of depression. For anxiety as indexed by the GAD-7 respectively both attenders ($M=10.06$) and non-attenders ($M=8.46$) fell into the moderate category.

Quality of Life: SF36

Significant differences were found between attenders ($M=21.87$) and non-attenders ($M=1.92$) in the psychological domain of quality of life (SF-36) on the physical health limitations subscale. An independent samples t-test revealed that those not attending the group reported higher scores on the limitations to physical health subscale of the SF36 (16.4) = -2.21 , $p<0.05$. This can be interpreted as attenders report feeling significantly less limited by their physical health problems than non-attenders

who scored lower on this subscale reflecting greater feelings of being limited as a result of physical health.

As the results from the other subscales did not reveal any significant differences (see table 5) this would suggest that attenders and non-attenders experienced a similar quality of life in other areas of their health-as indexed by the SF-36.

Health Beliefs: IPQ-B

There were significant differences between attenders (M=4.50) and non-attenders (M=2.23) with regards their self -reported levels on the IPQ-Brief personal control subscale. An independent t-test showed that those attending the group reported higher levels of control over their FND symptoms than those who did not attend $t(27) = -2.34, p < 0.05$. No other subscales differed significantly on the IPQ-B measure between attenders and non-attenders suggesting both types of participants held similar beliefs about other aspects of their FND.

Table 5 Descriptive and inferential statistics for psychological outcome measures for those attending and not attending the group intervention.

Psychological Domain	Outcome Measure	Attendees (n=16)			Non-Attendees (n=13)			Independent samples t-test		
		M	SD	Min-Max	M	SD	Min-Max	t	df	p
Cognition	Overall Score EPITRAK (age corrected)	32.07	6.20	17–42	30.08	6.77	19-39	-.81	26	0.42
Mood	PHQ9 Depression	14.50	7.81	3–27	12.08	5.87	2–23	-.93	27	0.36
	GAD7 Anxiety	10.06	6.70	2-21	8.46	7.02	1–21	-.63	27	0.54
Quality of Life	SF36 Physical Functioning	42.50	31.20	0-100	53.08	29.05	5-95	.94	27	0.36
	SF36 Physical health Limitations	21.87	35.21	0-100	1.92	6.93	0-25	-2.21	16.4	0.04
	SF36 Mental Health Limitations	39.56	45.91	0-100	56.46	47.90	0-100	.97	27	0.34
	SF36 Energy/Fatigue	26.56	20.31	0-85	30.00	16.07	15-75	.49	27	0.62
	SF36 Emotional Well Being	52.00	27.90	4-88	61.53	20.75	28-92	1.02	27	0.32
	SF36 Social Functioning	41.56	31.89	0-100	47.38	26.56	0-100	.53	27	0.60
	SF36 Pain	41.31	25.55	0-100	47.31	32.31	0-100	.56	27	0.58
	SF36 General Health	38.75	16.78	10-70	33.07	18.77	0-65	-.86	27	0.39
Health Beliefs	IPQ-Brief Consequences	6.75	2.69	0-10	6.61	1.75	4-10	-.15	27	0.88
	IPQ-Brief Timeline	8.44	1.86	5-10	8.53	2.06	5-10	.14	27	0.89
	IPQ-Brief Personal Control	4.50	3.01	0-10	2.23	1.96	0-5	-2.34	27	0.02
	IPQ-Brief Treatment Control	4.87	3.38	0-10	2.77	2.55	0-7	-1.85	27	0.07
	IPQ-Brief Identity	7.12	2.41	1-10	7.31	1.55	5-10	.24	27	0.82
	IPQ-Brief Illness Concern	5.81	3.20	0-10	6.92	2.10	3-10	1.07	27	0.29
	IPQ-Brief Coherence	3.69	3.20	0-10	4.54	2.92	1-10	.79	27	0.43
	IPQ-Brief Emotional Representation	6.12	3.36	0-10	6.08	2.50	2-10	-0.4	27	0.96
	IPQ-Brief Illness Threat	51.18	14.93	18-80	57.15	8.60	43-69	1.28	27	0.21

Healthcare Utilisation: Attenders and Non-Attenders

Services accessed by both attenders and non-attenders are reported at baseline in Table 6. In order to contextualise the different levels of healthcare participants would typically access, health sectors were divided into three sectors; Community care which comprised to GP and practice nurse appointments in primary care; Elective care which was comprised of specialisms regularly accessed by those with FND (e.g. Neurology, psychology, psychiatry and allied health professionals such as occupational therapy and physiotherapy. Outpatient, day patient and overnight stays were also detailed in the health sector. Non-elective care included visits to A+E; non-elective overnight admissions and treatment in intensive care.

Participants were asked to self-report their contact with these healthcare providers during the 3 months prior to their baseline appointment, responses were collected using the Client Services Receipt Inventory (see Table 6). Independent samples t-test revealed there were no significant differences between the total number of times attenders ($M=4.75$; $SD = 2.86$) and non-attenders ($M=3.56$; $SD = 3.47$) accessed community care services 3 months prior to baseline; $t(15) = -.77$; $p = .45$. Analysis on the total number of times elective care service were showed no differences between contact levels of attenders ($n= 12$; $M=5.50$; $SD=3.15$) and non-attenders ($n=8$; $M=4.00$; $SD = 3.12$) for the elective care sector; $t(18) = -.99$; $p= .34$. No comparative analysis was completed for non-elective services due to only 1 person accessing non-elective care in the non-attenders group making the sample size too small.

Table 6 Healthcare utilisation captured by the Client Services Receipt Inventory at baseline (T1) for those attending and not attending the group intervention

Health Sector	Provider	Attenders					Non-Attenders				
		n	%	Count	M	SD	n	%	Count	M	SD
Community Care	GP	7	24%	28	4.00	2.16	8	27%	31	3.88	3.56
	Practice Nurse	4	14%	10	2.50	2.38	1	4%	1	1.00	n/a
Elective Care	Neurology	2	7%	2	1.00	0.00	4	14%	5	1.25	.50
	Psychology	3	10%	11	3.67	2.08	0	0%	0	n/a	n/a
	Psychiatry	1	4%	8	8.00	n/a	3	10%	5	1.67	1.15
	Physiotherapy	4	14%	21	5.25	4.99	2	7%	9	4.50	2.12
	Occupational Therapy	2	7%	5	2.50	0.71	3	10%	3	1.00	.00
	Other	4	14%	11	1.25	0.74	4	14%	4	1.00	.00
	Elective Overnight stay	2	7%	3	1.50	0.71	0	0%	0	n/a	n/a
	Outpatient at Hospital	4	14%	7	1.25	0.50	1	4%	2	2.00	n/a
	Day patient at Hospital	1	4%	1	1.00	n/a	2	7%	2	1.00	1.00
Non-Elective Care	Accident and Emergency	2	7%	2	1.00	1.00	1	4%	1	1.00	n/a
	Non-Elective Overnight Stay	1	4%	1	1.00	n/a	0	0%	0	n/a	n/a
	Intensive Care	0	0%	0	n/a	n/a	0	0%	0	n/a	n/a

The unit and total costs of these patient contacts are reported in Table 7. As there were no significant differences between attenders and non-attenders in their healthcare utilisation costs are collapsed across attenders and non-attenders. In addition to the number of patients accessing the different health sectors, the number of times the service was accessed (count) was also detailed along with associated descriptive statistics. Costs used in Table 7 were taken from Public health Scotland cost books for NHS Scotland (<https://www.isdscotland.org/Health-Topics/Finance/Costs/Detailed-Tables/Speciality-Costs/>).

Community care costs were 17% of the total healthcare accessed, hospital based elective care services made up 80% of the cost total and non-elective emergency care was accessed by 10% of participants (£591), no participants accessed intensive care treatment. In total 29 participants with a diagnosis of FND accessed healthcare services 156 times over a three-month period with a total estimated cost of £26, 222.

Table 7 Healthcare costs based on responses from the 29 participants completing the Client Services Receipt Inventory at baseline (T1)

Health Sector	Health Care Provider	Number Accessing		Baseline Service Access (Previous 3 months)					Unit cost of service £	Total cost (count x unit cost) £	Mean cost per patient using (total cost/number patients accessing) £
		n	%	Count	Mean	SD	Min	Max			
Community Care	GP Contact	15	51%	59	3.93	2.89	1	12	70	4,130	275
	Practice Nurse	5	18%	11	2.20	2.17	1	6	43	473	95
Elective Care *	Neurology	6	21%	7	1.17	0.41	1	2	80	1,680	280
	Psychology	3	10%	11	3.67	2.08	2	6	97	1,067	355
	Psychiatry	4	14%	13	3.25	3.30	1	8	85	1,105	276
	Physiotherapy	6	21%	30	5.00	4.00	1	12	30	900	150
	Occupational Therapy	5	17%	8	1.60	0.89	1	3	86	688	138
	Elective Overnight Case ^a	2	7%	3	1.50	0.71	1	2	4,272	12,816	6408
	Outpatient at Hospital ^b	5	18%	7	1.40	0.55	1	2	230	1,610	322
	Day patient at hospital ^c	3	10%	3	1.00	0.00	1	1	332	996	332
	Accident and Emergency	3	10%	3	1.00	0.00	1	1	197	591	197
Non-Elective Care	Non-Elective Overnight Stay	1	4%	1	1.00	n/a	1	1	166	166	166
	Intensive Care	0	0%	n/a	n/a	n/a	n/a	n/a	-	-	-
Totals		-	-	156	-	-	-	-	-	26,222	452

* Other services category deleted as unable to provide cost information

^a Individual specialities not captured cost reflects generic gross cost per case for elective inpatient for NHSG from Public Health Scotland book R044X

^b Individual specialities not captured cost reflects generic speciality outpatient gross cost for NHSG from Public Health Scotland book R044X

^c Individual specialities not captured cost reflects generic speciality day patient gross cost for NHSG from Public Health Scotland book R044X

HRQoL EQ-5D-5L: Attenders and Non-Attenders

As non-attenders did not complete the group intervention, data from the EQ-5D-5L was only collected at baseline (T1). As there was no comparison data, it was not possible to calculate difference in quality adjusted life years. However, the data were analysed to explore if there were significant differences in the pattern of responses between attenders and non-attenders for the HRQoL dimensions in the EQ-5D-5L.

Participants chose the response that best reflected their health in five quality of life domains, on the day the questionnaire was completed. A summary of the distribution of frequency data for the EQ-5D-5L dimensions for both attenders and non-attenders at baseline (T1) are shown in Table 8. Fishers Exact test was used to analyse the frequency counts of the responses on the EQ-5D-5L due to low expected counts in the comparisons. No statistical differences were found between those attending or not attending the group intervention which suggested that none of the dimensions in the EQ-5D-5L included were related to whether the participant attended the intervention or not.

The mean of the EQ-visual analogue scale (EQ-VAS) scores where the participant rated their health on the day between 0 is worst health imaginable and 100 is the best health imaginable. for both attenders and non-attenders were analysed using an independent samples t-test. No significant differences were found between the VAS ratings for attenders (M=57.25; SD =23.11) and non-attenders (M=63.08; SD = 15.48) at baseline; $t(27) = 0.77$; $p = 0.44$ suggesting these factors did not influence attendance.

Table 8 EQ-5D-5L frequencies and percentage reported by dimension and level for group attenders and non-attenders at baseline (T1)

	Attenders (n=16)	Non- Attenders (n=13)	Fishers Exact test P (two-tailed)¹
Mobility (%)			0.71
1 No problems – 5 Unable to walk			
Level 1	4 (25%)	4 (31%)	
Level 2	5 (31%)	6 (47%)	
Level 3	5 (31%)	3 (22%)	
Level 4	2 (13%)	0 (0%)	
Level 5	0 (0%)	0 (0%)	
Self-Care (%)			0.28
1 No problems – 5 Unable to wash/dress			
Level 1	5 (31%)	6 (47%)	
Level 2	9 (57%)	5 (38%)	
Level 3	0 (0%)	2 (15%)	
Level 4	1 (6%)	0 (0%)	
Level 5	1 (6%)	0 (0%)	
Usual Activities (%)			0.87
1= No problems – 5 Unable to do usual activities			
Level 1	3 (19%)	2 (15%)	
Level 2	6 (37%)	5 (38%)	
Level 3	4 (25%)	5 (38%)	
Level 4	1 (6%)	1 (9%)	
Level 5	2 (13%)	0 (0%)	
Pain Discomfort			0.38
1 No pain – 5 Extreme Pain			
Level 1	1 (6%)	3 (22%)	
Level 2	5 (31%)	1 (9%)	
Level 3	7 (44%)	6 (47%)	
Level 4	3 (19%)	3 (22%)	
Level 5	0 (0%)	0 (0%)	
Anxiety/Depression			1.00
1 Not anxious/depressed – 5 Extremely Anxious/depressed			
Level 1	6 (37%)	6 (47%)	
Level 2	4 (25%)	3 (22%)	
Level 3	3 (19%)	3 (22%)	
Level 4	2 (13%)	1 (9%)	
Level 5	1 (6%)	0 (0%)	

Study Aim 2

The second aim of the study was to identify what changes occurred in the standardised outcome measures indexing psychological constructs, for those completing the group intervention.

Group Completers Psychological Outcome Measures

Cognitive Function: Epiritak

A repeated measures ANOVA showed that the level of performance for cognitive function, as indexed by the Epiritak, remained unchanged for completers between T1 (M=33.20) and T4 (M=32.70) with no significant differences found; [$F(1,9) = 0.12$; $p = 0.73$]. This result suggested participants cognitive profile did not change significantly between baseline and follow up.

Mood: Depression PHQ-9 & GAD-7

A repeated measures ANOVA was conducted to explore symptoms of depression and anxiety across the four time points, which showed no changes in scores as indexed by the PHQ9 [$F(3, 30) = 2.42$; $p = 0.86$] or GAD7 ; [$F(3, 30) = 1.01$; $p = 0.40$]. (See **Error! Reference source not found.9**)

Quality of Life: SF36

The energy/fatigue subscale of the SF-36, showed a significant improvement for completers at T4 (see **Error! Reference source not found.9**). A repeated measures ANOVA indicated that scores for the energy/fatigue subscale differed across the four data timepoints, $F(3,27) = 3.22$ $p < 0.05$. Inspection of pairwise comparisons revealed that post-group scores (T3; M=14.00) differed significantly from follow up (T4; M=30.00) $p < 0.05$ with completers reporting higher levels of energy/lower levels of fatigue at follow-up compared to post-group scores. Findings for changes in the SF-36

subscale suggest that although there were no significant differences during the intervention phase fatigue levels were reported as significantly less between the intervention ending and follow up.

Health Beliefs: IPQ-B

The repeated measures ANOVA for the coherence subscale on the IPQ-B (see **Error! Reference source not found.**9). revealed significant revealed that scores for the coherence subscale differed significantly across the four study time points, $F(3, 30) = 5.67$; $p < 0.00$. Inspection of pairwise comparisons revealed that the pre-intervention scores [Baseline (T1; $M = 4$) and Pre-group (T2; $M = 4$)] did not differ significantly. There were also no significant differences between post-intervention scores [Post-Group (T3; $M = 7.36$) and Follow up (T4; $M = 6.27$)]. However, there were significant differences between pre and post intervention scores with significant differences between T1 and T3 ($p < 0.01$) also T1 and T4 ($p < 0.05$). T2 scores differed significantly with T3 ($p < 0.01$) and T4 ($P < 0.05$). The results found in the IPQ-B coherence subscale suggest that participants rated their understanding of their illness highest immediately after the group at T3 and this increase in understanding was still significant, although reduced slightly, 3 months later at T4.

Table 9 Group Completers performance on Psychological Outcome Measures

Psychological Domain	Outcome Measure	T1		T2		T3		T4		Repeated Measures ANOVA		
		M	SD	M	SD	M	SD	M	SD	F	df	Sig
Cognition	Overall Score EPITRAK (age corrected)	33.20	5.18	Not Administered				32.70	7.08	0.12	1, 9	0.73
Mood	PHQ9 Depression	16.27	7.32	17.27	7.95	15.64	6.8	13.45	6.44	2.42	3, 30	0.86
	GAD7 Anxiety	10.73	6.97	11.00	7.54	11.18	7.45	8.73	6.67	1.01	3, 30	0.40
Quality of Life	SF36 Physical Functioning	42.50	32.34	61.50	34.88	63.50	35.67	52.00	35.21	1.41	3, 27	0.26
	SF36 Physical Health Limitations	15.00	33.75	5.00	10.54	17.50	28.99	20.00	32.91	0.79	3, 27	0.51
	SF36 Mental Health Limitations	30.00	48.30	33.33	47.14	50.00	52.70	50.00	45.13	1.95	3, 27	0.18
	SF36 Energy/Fatigue	19.00	13.08	18.75	17.92	14.00	19.83	30.00	23.57	3.22	3, 27	0.04
	SF36 Emotional Well Being	47.60	28.00	45.50	29.29	38.40	30.18	51.60	26.83	0.98	3, 27	0.42
	SF36 Social Functioning	32.60	27.10	31.25	27.80	27.50	22.67	43.85	21.29	2.50	3, 27	0.81
	SF36 Pain	35.90	23.24	26.75	23.04	35.00	29.11	37.50	26.33	2.71	3, 27	0.06
	SF36 General Health	37.50	18.45	31.00	21.45	24.5	20.88	33.5	20.42	1.58	3, 27	0.22
Health Beliefs	IPQ-Brief Consequences	7.64	1.80	7.55	1.81	7.45	2.11	6.91	1.87	0.81	3,30	0.50
	IPQ-Brief Timeline	9.00	1.41	8.82	1.66	8.27	2.10	8.73	1.85	1.14	3,30	0.35
	IPQ-Brief Personal Control	4.18	3.12	3.73	2.76	4.82	2.52	3.73	2.33	0.61	3,30	0.61
	IPQ-Brief Treatment Control	4.09	3.56	3.36	2.94	4.09	3.11	3.36	2.87	0.39	3, 30	0.75
	IPQ-Brief Identity	7.91	1.58	7.55	1.92	8.09	1.04	7.64	1.91	0.48	3, 30	0.70
	IPQ-Brief Illness Concern	6.73	2.72	6.91	2.63	6.64	2.87	6.09	2.43	0.58	3,30	0.63
	IPQ-Brief Coherence	4.00	3.29	4.00	2.61	7.36	1.21	6.27	3.04	5.67	3,30	0.00
	IPQ-Brief Emotional Representation	6.73	2.87	6.73	3.04	6.73	3.10	6.82	3.06	0.12	3, 30	0.99
	IPQ-Brief Illness Threat	55.55	12.36	56.73	14.77	50.91	10.62	52.82	13.30	1.21	3, 30	0.32

Study Aim 3

The third aim of this research compared the healthcare utilisation and associated healthcare costs for those attending the group intervention between T1 and T4.

Group Completers Healthcare Utilisation: CSRI

Services accessed by completers at both baseline (T1) and follow up (T4) are reported in Table 10. Health sectors were, as previously, divided into three sectors; Community care (GP and practice nurse appointments in primary care); Elective care (e.g. Neurology, psychology, psychiatry and allied health professionals. Outpatient, day patient and overnight stay) and Non-elective care included (A+E; non-elective overnight admissions and intensive care). Completers self-reported their contact with each healthcare providers during the 3 months prior to their baseline appointment, and 3 months prior to their follow up appointment.

A paired sample t-test revealed there were no significant differences between the total number of times completers accessed community care health services 3 months prior to baseline (T1) ($M=7.12$; $SD=5.81$) and follow up (T4) ($M=3.62$; $SD=3.20$); $t(7) = 1.61$; $p = .15$. No significant differences were found for number of elective healthcare contacts at T1 ($n=7$; $M=6.86$; $SD=3.13$) and T4 ($n=7$; $M=7.57$; $SD=7.82$); $t(6) = -0.25$; $p = .81$. Paired samples t-tests revealed no significant differences between non-elective care contacts at baseline ($n=2$; $M=1.50$) and follow-up (T4) ($n=2$; $M=2.00$); $t(1) = 1$; $p = .50$.

The unit and total costs of healthcare contacts for completers are reported in table 11. There was a reduction of costs related to community and elective care. However for non-elective care there were no differences found for costs between T4 and T1.

Table 10 Completers Healthcare Utilisation captured by the CSRI for baseline (T1) and Follow up (T4)

Health Sector	Provider	Completers T1					Completers T4				
		n	%	Count	M	SD	n	%	Count	M	SD
Community Care	GP	5	45	24	4.80	1.92	6	54	21	3.50	1.76
	Practice Nurse	3	27	9	3.00	3.07	7	64	11	1.57	0.79
Elective Care	Neurology	1	9	1	1.00	n/a	1	9	1	1.00	n/a
	Psychology	3	27	11	3.67	2.08	1	9	4	4.00	n/a
	Psychiatry	1	9	8	8.00	n/a	2	18	9	4.50	4.95
	Physiotherapy	2	18	14	7.00	7.14	2	18	10	5.00	1.41
	Occupational Therapy	2	18	5	2.50	0.71	2	18	5	2.50	2.12
	Other	2	18	3	1.50	0.71	4	36	11	2.75	0.96
	Elective Overnight stay	2	18	3	1.50	0.71	0	0	0	n/a	n/a
	Outpatient at Hospital	2	18	2	1.00	0.00	3	27	10	3.33	3.21
	Day patient at Hospital	1	9	1	1.00	n/a	0	0	0	n/a	n/a
Non-Elective Care	Accident and Emergency	2	18	2	1.00	0.00	2	18	2	1.00	0.00
	Non-Elective Overnight Stay	1	9	1	1.00	n/a	2	18	2	1.00	0.00
	Intensive Care	0	0	0	n/a	n/a	0	0	0	n/a	n/a

Table 11 Healthcare costs comparison at T1 and T4 for those completing the intervention

Health Sector	Provider	Unit Cost £	Baseline (T1)				Follow Up (T4)				Cost Change £ (T4-T1)	Cost Change £ (per sector)
			n	n%	Count	Total cost (count x unit cost	n	n%	Count	Total cost £ (count x unit cost		
Community Care	GP Contact	70	5	45	24	1,680	6	54	21	1,470	-210	-124
	Practice Nurse	43	3	27	9	387	7	64	11	473	86	
Elective Care *	Neurology	80	1	9	1	80	1	9	1	80	0	-12,022
	Psychology	97	3	27	11	1,067	1	9	4	388	-679	
	Psychiatry	85	1	9	8	680	2	18	9	765	85	
	Physiotherapy	30	2	18	14	420	2	18	10	300	-120	
	Occupational Therapy	86	2	18	5	430	2	18	5	430	0	
	Elective Overnight Case ^a	4,272	2	18	3	12,816	0	0	0	0	-12, 816	
	Outpatient at Hospital ^b	230	2	18	2	460	3	27	10	2,300	1,840	
	Day patient at hospital ^c	332	1	9	1	332	0	0	0	0	-332	
Non-Elective Care	Accident and Emergency	197	2	18	2	394	2	18	2	394	0	166
	Non-Elective Overnight Stay	166	1	9	1	166	2	18	2	332	166	
	Intensive Care	0	0	0	0	n/a	0	0	0	0	0	
Totals		-	-	-	81	-	-	-	75	-	-11,980	-11, 980

*Other services category deleted as unable to provide cost information

^a Individual specialities not captured cost reflects generic gross cost per case for elective inpatient for NHSG from Public Health Scotland book R044X

^b Individual specialities not captured cost reflects generic speciality outpatient gross cost for NHSG from Public Health Scotland book R044X

^c Individual specialities not captured cost reflects generic speciality day patient gross cost for NHSG from Public Health Scotland book R044X

Group Completers HRQoL Cost Utility Analysis: EQ-5D-5L

The frequency data at baseline (T1) and follow up (T4) were analysed to explore if there were differences in the pattern of responses given by group completers at T4 compared to T1 (see Table 12). At domain level, the number of participants experiencing problems reduced over for mobility, self-care and usual activities. There were no changes in the number of people reporting problems for the domains of pain/discomfort and anxiety/depression. However, the pattern of data from T4 from T1 did highlight that there were changes at participant level occurring on the dimensions of the EQ-5D-5L. For example, at T4 no participants were reporting symptoms at Level 5 whereas at T1 level 5 scores were reported for self-care usual activities and anxiety/depression.

In order to explore changes at participant level more fully, the changes in the distribution of the domain levels for each participant were calculated using the Paretian classification of health change (see Devlin, Parkin & Browne, 2010). The T4 and T1 health profiles were considered using four possible outcomes (See Table 13)

1. There was no change in health profile at T4 from T1 as indexed by the EQ-5D-5L
2. T4 health profile is better than T1 indicating health as improved according to the EQ-5D-5L
3. The T4 health profile is poorer than T1 indicating poorer health at follow up as measured by the EQ-5D-5L.
4. It is not possible to compare the health profiles at T4 and T1, further information would be required prior to categorising changes as an being improved or worsened.

Table 12 EQ-5D-5L frequencies and percentage reported by dimension and level for group completers at baseline (T1) and follow up (T4)

	Mobility				Self-Care				Usual Activities				Pain/discomfort				Anxiety/Depression			
	T1 Freq	T1 %	T4 Freq	T4 %	T1 Freq	T1 %	T4 Freq	T4 %	T1 Freq	T1 %	T4 Freq	T4 %	T1 Freq	T1 %	T4 Freq	T4 %	T1 Freq	T1 %	T4 Freq	T4 %
Level 1	3	28%	4	36%	3	27%	5	46%	0	0%	1	9%	0	0%	0	0%	4	36%	4	36%
Level 2	4	36%	3	28%	6	55%	4	36%	5	45%	6	55%	4	36%	3	27%	3	27%	3	27%
Level 3	2	18%	1	9%	0	0%	1	9%	4	36%	3	27%	5	45%	5	46%	2	18%	2	18%
Level 4	2	18%	3	27%	1	9%	1	9%	0	0%	1	9%	2	18%	3	27%	1	9%	2	18%
Level 5	0	0%	0	0%	1	9%	0	0%	2	18%	0	0%	0	0%	0	0%	1	9%	0	0%
Total	11	100%	11	100%	11	100%	11	100%	11	100%	11	100%	11	100%	11	100%	11	100%	11	100%
Number reporting some problems	8 (73%)		7 (64%)		8 (73%)		6 (55%)		11 (100%)		10 (91%)		11 (100%)		11 (100%)		7 (64%)		7 (64%)	
Change in number reporting problems	-1				-2				-1				0				0			

Table 13 Changes in health at participant level from baseline (T1) to Follow up (T4) for the EQ-5D-5L dimensions as categorised by the Paretian Classification of health change

	Mobility	Self-Care	Usual Activities	Pain/discomfort	Anxiety/Depression	T4-T1 Health State
No change	0	0	0	0	0	1
Improved	1	4	5	2	5	7
Worse	1	0	1	1	2	3
Mixed Change	0	0	0	0	0	0
Total	2	4	6	3	7	11

Seven patients reported improvements in their overall health profile at T4 compared to T1. The greatest improvements were seen in usual activities (n=5), anxiety and depression (n=5) and self-care (n=4), pain (n=2) and mobility (n=1) were also reported as improved at T4 by participants. Three participants rated their health state as poorer at T4. Mood (n=2), (mobility (n=1), usual activities (n=1) and pain/discomfort (n=1) showing higher ratings at T4 than T1. One participant recorded no change in scores on the EQ-5D-5L from T1 to T4.

Due to the small number of people completing the intervention, there was an insufficient number of completers (n=11) to allow utility values to be calculated for the EQ-5D-5L. This means it was not possible to calculate differences in quality adjusted life years for this current intervention at this time. However, the pattern of results noted in the measure suggests the EQ-5D-5L can be successfully administered in this population.

A paired samples t-test was used to explore if the scores on the EQ-visual analogue scale (EQ-VAS) at T4 differed from T1 for those participants completing the intervention. No significant differences were found between T4 (M=55.09; SD =28.32) and T1 (M=53.27; SD = 24.97); $t(10) = -0.29$; $p = 0.77$.

Study Aim 4

The final aim of the study looked at the experience of the participants to determine if the experiential aspect of the group met their expectations and significantly improve the subjective levels of knowledge about FND post intervention.

Group Completers Experiential Measures

Expectations of the participants

Full details of the expectation's participants reported about taking part in the group can be found in Appendix 8. The following themes were found within the participant pre-group expectations (see Table 14)

Table 14 Theme of participant expectations for the group intervention

Theme of Expectation	N (max = 11)
Increase knowledge of FND	10
Access peer support	8
Managing symptoms	5
Informing future healthcare for both professionals and patients	4

Most participants hoped to increase their knowledge on FND and meet others with the condition to share and learn from each other's experiences. In addition to learning how to manage symptoms, it was important for some of those attending to help not only others receiving a diagnosis but also to help educate healthcare professionals. To help those working with people with FND to be more empathic to the condition in order to reduce patients potentially feeling 'fobbed off' (see patient 2 comments in Appendix 9)

Subjective Knowledge of Completers

Significant differences were found for all knowledge objectives between T2 (Pre) and T3 (Post) group (see Table 15). Paired samples t-test revealed that participants rated their knowledge on what FND is higher at T3 (M=4.38) than T2 (M = 3.46); $t(12) = -3.49$; $p < 0.00$. Knowledge on how FND was diagnosed also increased significantly for completers between T2 (M=2.92) and T3 (M=4.08); $t(12) = -4.21$; $p < 0.00$. Understanding of how physical and psychological processes interact in their body was rated as higher at T3 (M=4.15) than T2 (M=3.54); $t(12) = -2.31$; $p < 0.05$. It was reported more was known about coping strategies at T3 (M=4.31) than T2 (M=3.08); $t(12) = -3.12$, $p < 0.01$. Participants reported they knew more about what made their symptoms worse at T3 (M=4.46) than T2 (M=3.46); $t(12) = -2.94$, $p < 0.01$. Knowledge on useful resources also increased significantly between T2 (M=2.54) and T3 (M=4.23); $t(12) = -5.50$, $p < 0.00$. There were no significant difference found, $t(12) = -0.94$, $p = 0.37$ between T2 (M= 4.62) and T3 (M=4.92) on how helpful it would be to meet others as there was a ceiling effect with the majority of participants rating they would find meeting others very useful both at T2 and again at T3.

Table 15 Self-reported knowledge levels of completers at T2 (pre) and T3 (post) group intervention

	Pre-Group Knowledge (n=13)			Post Group Knowledge (n=13)			Paired Samples t-test (df=12)	
	M	SD	Min- Max	M	SD	Min- Max	t	p
What FND is	3.46	0.97	2-5	4.38	.51	4-5	-3.49	.00
Diagnosing FND	2.92	1.04	1-5	4.08	.64	3-5	-4.21	.00
How Physical & Psychological Process interact in the body	3.54	0.88	2-5	4.15	.69	3-5	-2.31	.04
Coping Strategies	3.08	1.32	1-5	4.31	.48	4-5	-3.12	.01
Why symptoms get worse	3.46	1.27	1-5	4.46	.52	4-5	-2.94	.01
What resources are available	2.54	0.97	1-4	4.23	.60	3-5	-5.50	.00
How helpful to meet others	4.62	1.12	1-5	4.92	.28	4-5	-0.94	.37

Experience of the intervention

At the end of the second group, participants were invited to complete an anonymous group evaluation form with space to leave comments if desired (See appendix 9 for summary of qualitative comments). 85% of participants (n=11) opted to do see figure 3.

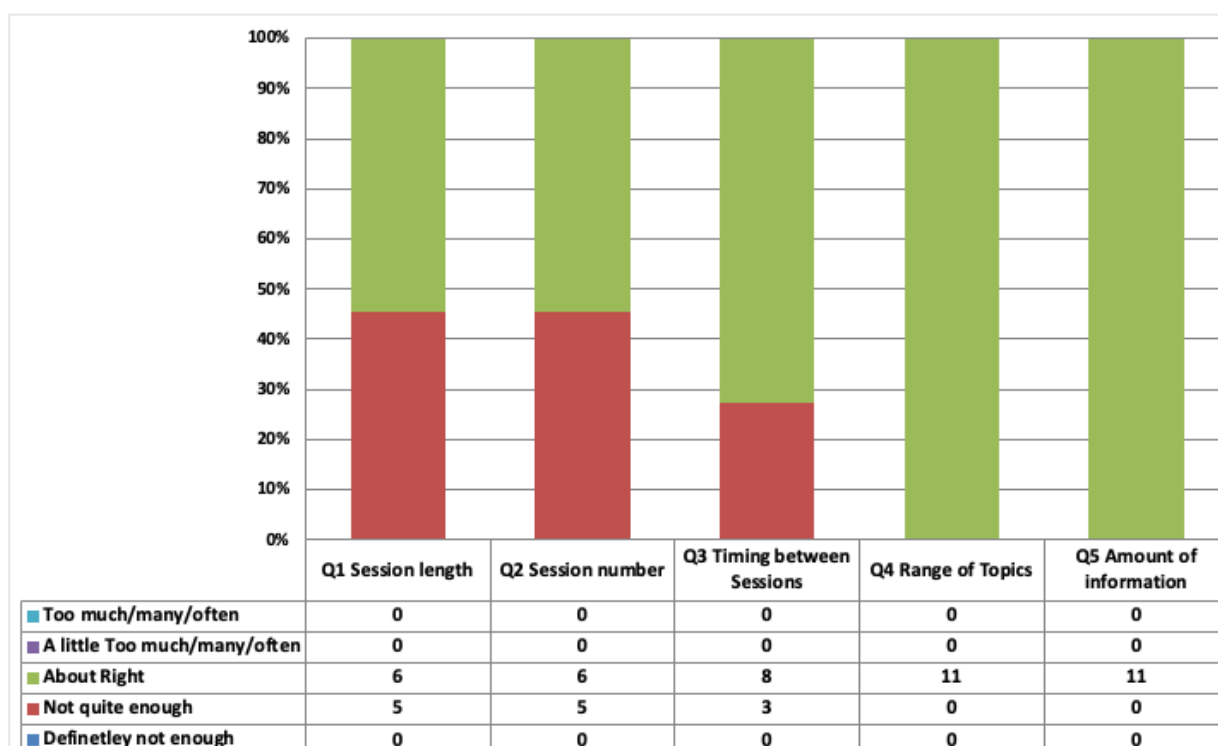


Figure 3 Participant evaluation of group intervention sessions

With regards the session length, 4 participants felt the sessions could be slightly longer at 2 hours rather than the 1.5 hour in the study. This extra time was felt to allow more time for questions and group discussion. 45% (n=5) felt there could be an additional session to consolidate info and give opportunity to meet the others again to learn from each other. 27% (n=3) participants stated a preference for the sessions to be a week apart but the majority felt the fortnightly format was about right.

All participants felt the range of topics and the amount of information presented on each one was about right. One participant felt more time could be spent on each topic, another felt there was a lot to take in. The volume of information was reiterated by another participant who felt that weekly sessions over 4-6 weeks would give participants more time to consolidate information and discuss with the group the following week.

All participants found the room suitable and the group facilitators approachable (see figure 4). One participant noted the helpfulness of the neurologist's information videos and the relaxed atmosphere in the session. Although everyone found the group information accessible a few participants stated concerns about their ability to remember the information later due to the difficulties they were experiencing with their memory at the time of the group.

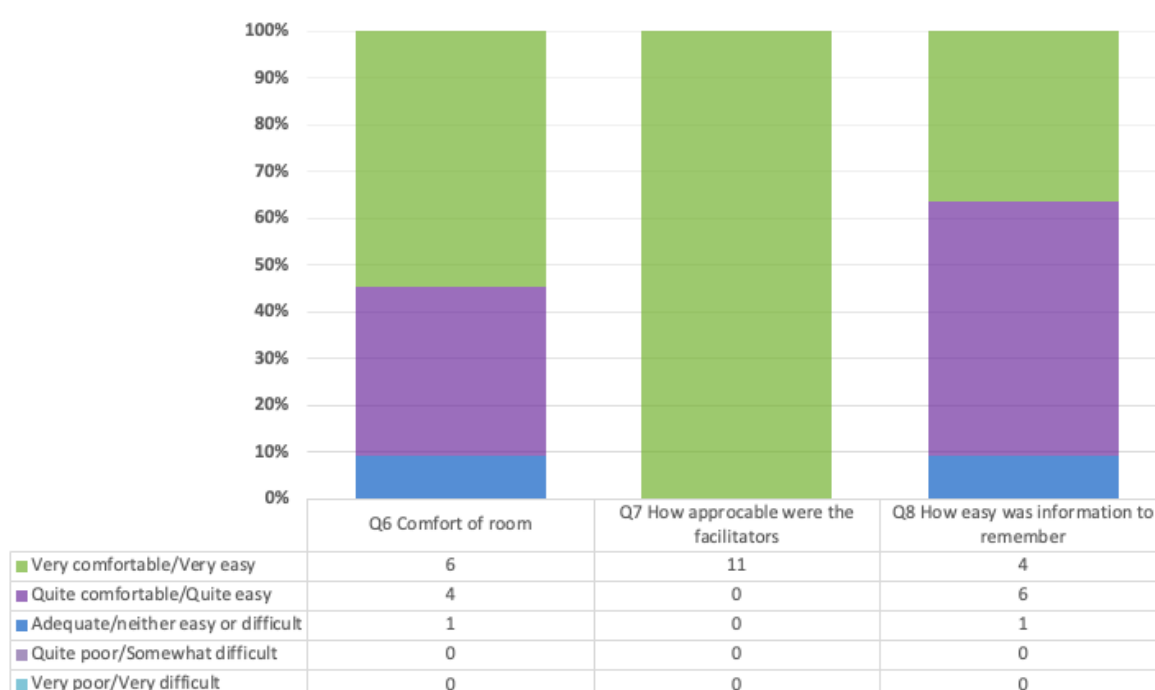


Figure 4 Participant rating of group delivery variables

The elements of the intervention participants particularly liked were the friendly atmosphere and having a mix of peers and professionals. One participant reflected they felt the group normalised FND and brought awareness to the fact that some healthcare professionals do not know much about it. Changes that were suggested mainly included more time for peer discussion and some more time to digest the information. All of those completing the group would recommend it to a friend. Further comments were related to knowing more about FND and taking away the feeling of being alone after diagnosis. Signposting to other resources was also considered helpful. There was also mention that the group might be helpful to families supporting those with FND and not just the patients. A summary of these qualitative comments can be seen in Appendix 9.

Discussion

This purpose of this research was to develop a brief intervention and evaluate the its feasibility in supporting people with FND within NHS Grampian. This study aims to address a current gap in local service provision. Data were collected over four timepoints; baseline (T1), pre-group (T2), post-group (T3) and 3 months after the group, follow-up (T4).

There were no significant differences in demographic variables between attenders and non-attenders. Significant differences were found for psychological outcomes with those choosing not to attend the first session rating their physical health limitations as significantly greater than those who attended. In addition non-attenders reported lower levels of perceived personal control than attenders. Follow up data revealed those completing the group reported significantly reduced fatigue at follow up. In addition 63% of completers reported improved health status at follow-up on health-related quality of life.. Subjective levels of self-report knowledge about FND increased significantly from pre to post intervention. Although their understanding of FND was reported highest post intervention this increased knowledge level remained in follow up data. Some participants thought two sessions was enough but with some preferring some more, thoughts around duration were also mixed. However the topics included and amount of information was about right. Participants found the information was easy to remember and felt comfortable with the room and the facilitators.

In the current study, there was an attrition rate of 45% (n=13) for participants who attended the baseline session but did not attend the group intervention. This was

an identical rate of non-attendance found in a one to one treatment programme for FND previously (Demartini, Batla, Petrochilos, Fisher, Edwards et al., 2014).

Demographic variables in the current research were compared and revealed no significant differences between attenders and non-attenders. The high prevalence of females in the study match gender and age profiles of people with FND found previously (see Carson & Lehn, 2016; Edwards & Bhatia, 2012 for reviews). This is the first study to have included a measure of health inequalities in an FND population taking part in a therapeutic intervention. Although no significant differences were found in the distribution of participants across the SIMD quintiles, the majority of participants resided in the higher socio-economic areas. These figures should be interpreted in the context of location with this finding likely to have reflected the lower distribution of people living in these locations within the Grampian area as a whole rather than being an indication of health inequalities within the study sample

The analysis of data measuring psychological constructs showed mixed results. There were no significant differences between types of participant for the cognitive task or for either measures of mood. However, non-attenders reported significantly lower levels of personal control at baseline than those who went on to attend the intervention. Those not attending the intervention also rated themselves as being significantly more limited in their life by physical health.

The personal control subscale within the IPQ-Brief has been previously found to correlate with a measure of self-efficacy and as a result, may be a proxy indicator for health beliefs indexed by this subscale (Broadbent et al., 2006). Health beliefs are formed and maintained by the ongoing appraisal of symptoms and how successfully these are managed (Leventhal, Halm, Horowitz, Leventhal, Ozakinci et al., 2004).

Interpreting the finding that non-attenders perceive low personal control and feel highly limited physically would suggest that a lack of confidence in personal ability to manage symptoms becomes a self-fulfilling prophecy which goes on to affect physical function resulting in a lower quality of life. A comparable pattern of results has been found using similar measures in a population with chronic pain previously (de Rooji, DeBoer, van der Leeden, Steultjens & Dekker, 2014).

In previous research, perceived levels of personal control have been associated with an anticipation that the proposed treatment would not be effective (van Wilgen, van Ittersum, Kaptein, & van Wijhe, 2008). A non-significant trend in the current data potentially supported this finding in the current data set with the treatment control subscale of the IPQ-brief also being reported lower by non-attenders than attenders. This suggested that participants who did not take part were more likely to believe the intervention would not help to manage their symptoms. Although this difference between the attenders and non-attenders on the treatment control subscale did not reach significance, the probability value approached a level of significance which suggested the result may be attributable to insufficient power in the study.

To maximise engagement with interventions there needs to be an awareness of how best to address maladaptive beliefs that participants with FND may have that could lead to self-imposed barriers to accessing treatment. Those with FND have shown evidence of bias in their thinking previously by deciding on the likelihood of a particular outcome long before all the information was available for evaluation (Pareés, Kassavetis, Saifee, Sadnicka, Bhatia, et al., 2012). This cognitive bias in the context of health beliefs may indicate that some people with FND update their mental model of their condition in a similar impulsive manner resulting in maladaptive beliefs regarding

their symptoms (Pareés et al., 2012) which may subsequently link to impact negatively on their beliefs about their ability to cope.

The current intervention was facilitated by a Consultant Clinical Neuropsychologist and a trainee psychologist. The link the current intervention had with mental health may potentially contributed to non-attendance. There is historical evidence to suggest that engaging with mental health services can be problematic for people with FND. A lack of engagement may be as a result of experiencing feelings of stigma after being diagnosed with FND (Rommelfanger, Factor, LaRoche, Rosen, Young & Rapaport, 2017). Receiving information that psychological factors can explain the physical symptoms being experienced may be difficult to reconcile (Rommelfanger et al., 2017). The processing of the diagnostic label of illness and the meaning a person extracts from this has been identified as important in the self-regulation model of health beliefs (Leventhal et al., 1980; 1984).

There were no significant differences in the healthcare utilisation patterns between attenders and non-attenders in the current data. However, current results add to evidence that the health care utilisation of people with FND involved a high number of services being accessed on a regular basis. It is important to interpret these findings with caution as these figures were self-report and not corroborated with health records to verify accuracy. As a result these figures may be an under representation of clinical contacts. This result showed primary care were the most frequently used services by those with FND in the current sample.

Due to only completing baseline data it was not possible to conduct quality adjusted life years for attenders and no-attenders. There were no significant differences between attenders and non-attenders on the frequency data for current levels of function

over the 5 levels of quality of life or for quality of life reported on the visual analogue scale.

Previous research has included an attempt to differentiate between those attending or not attending an FND therapeutic intervention previously (Goldstein et al., 2004). The previous research comparison was limited as it focused on demographic and symptom profiles only and did not include a statistical analysis (Goldstein et al., 2004). The current research is the first to widen the comparison. Inferential statistics were used to explore demographic, psychological and healthcare utilisation variables to ascertain if there were significant differences between those with FND who attended and did not attend the group intervention.

The second aim considered potential changes in psychological measures for those completing the group intervention over the four data collection timepoints of the intervention. No significant differences were found for cognitive performance, participants fell into the average performance range and this was stable between baseline and follow up. This result suggests there may be inconsistencies between objective and subjective experience of patients with FND regarding their cognitive abilities. This is the first study to have included a measure of cognitive function in the evaluation of a psychological intervention. Although there is limited research on the cognitive profile of those with FND, existing research has interpreted the discrepancy between subjective cognitive performance and objective measures may reflect a lack of attention capacity (Teodoro et al., 2018). Attentional resources may be utilised with internal monitoring of bodily symptoms in addition to dealing with pain and fatigue (Teodoro et al., 2018).

A strength of the current study was the collection of data over four time points. The inclusion of a baseline and follow up period allowed natural fluctuations over time to be included rather than focusing in on pre and post measures only which would have been over a short duration of 2 weeks. Scores on the mood measures also did not change significantly over the four data collection time points, however, there appeared to be a natural fluctuation of symptoms over time. The symptoms of depression and anxiety are accommodated by the SRM of health beliefs which suggests that these symptoms are the by-product of symptoms continuing despite attempts to manage them (Leventhal et al., 2007).

There were significant differences found in the quality of life measure with the subscale of energy/fatigue significantly improved from post group. This was interesting as this occurred despite no change in cognition, mood, areas that have been found to impact on quality of life for those with FND previously (Vechetova et al., 2018). This may reflect the benefit of allowing a period of consolidation in therapeutic interventions to allow participants time to incorporate techniques in their daily routine.

In addition, there were significant improvements in the health belief measure for illness understanding (coherence). The significant difference found post group remained significant at 3 month follow up. Cope et al., (2017) also found improvements on the coherence subscale for their brief intervention, however they did not include a follow up period and their sample included only patients with Functional non-epileptic attacks. The current study extends these findings to a mixed FND sample.

Healthcare utilisation and healthcare costs were also explored for those completing the group intervention. No significant difference was found in the number of healthcare contacts between T1 and T4. Care should be taken when interpreting this finding as the data are based on subjective self-report which has not been verified with health records.

Healthcare utilisation and healthcare costs were also explored for those completing the group intervention. Similar to findings between attenders and non-attenders there were no significant findings from the number of contacts at T1 and T4 however similar to the analysis of attender and non-attenders the data analysed was self-reported and not verified with health records which could result in an under reporting of contacts.

Although limited numbers meant QALY's were unable to be calculated for the current intervention, the data from the EQ-5D-5L showed that some of the participants moved from having problems to no problems on the mobility, self-care and usual activities dimensions. In addition, improvement in overall health state was reported by 63% of the completers. This is an encouraging result from this exploratory work and fits with the findings of lower fatigue and increased illness coherence reported earlier from the psychological measures. In order to explore the robustness of this finding a larger sample would be required than is in the current study. Seven patients reported improvements in their overall health profile at T4 compared to T1.

Measures of subjective participant knowledge of FND and a questionnaire capturing the participants experience of the group were included in order to gain feedback to inform future versions of the intervention. Results showed that participants wanted to increase their knowledge of FND, access peer support, manage symptoms

and help inform future healthcare for patients and healthcare providers. They felt the group helped them meet these expectations. Subjective levels of knowledge about FND increased significantly between pre and post group for all study aims. Constructing a narrative from the additional comments it became apparent that the group format addressed feeling isolation meaning participants felt validated and less alone. This feeling of isolation is common in those with FND (Nettleton, Watt, O'Malley & Duffey, 2004). Reduced isolation as a result of meeting others has been found in previous group interventions exploring patient feedback previously (Cope et al., 2017; Blake, Ablitt, Ruffman, Morley, William et al., 2019). It was encouraging to see that participants were unanimous in rating the topics included as acceptable with the amount of information. The current study based the intervention on a biopsychosocial model allowing a holistic approach to be taken rather than positioning symptoms as a result of an arbitrary physical/mental health dichotomy when trying to explain the diagnosis.

The use of video commentary from the neurologists was also received well. Developing this resource may have increased the validation felt by participants and it addressed concerns such as misdiagnosis. Many of the participants had voiced concerns around the FND diagnosis and had interpreted the process of as reflecting uncertainty around the diagnosis rather than as a corroboration of patterns that relate to a functional disorder. This highlighted the current methods of diagnosis by elimination that may contribute to the high healthcare utilisation patterns seen in the FND population. Session length and number of sessions were indecisive with some thinking 2 was enough some preferring some more, some felt timing between session could have been longer, however and amount of information was about right.

Participants found the information was easy to remember and felt comfortable with the room and the facilitators.

Strengths and Limitations

The current research showed several strengths in the methodology. Incorporating an analysis between attenders and non-attenders allowed a comparison of these participants on a range of data. There were potential differences identified around health beliefs and quality of life revealed from this analysis. A wide range of outcome measures based on relevant clinical guidelines were completed with a mix of FND subtypes which allowed generalisation across the wider FND population. Including a baseline and follow up period allowed results to be considered over a longer temporal duration. Follow up data showed that effects found in participants appear to sustain post intervention.

This research was innovative and well received by NHS staff and those with FND. There was a lot of interest from other disciplines regarding when the intervention would be clinically available in order for them to be able to refer patients for treatment. The intervention was well supported within the neurology department who were keen to support recruitment. It is hoped that the intervention will be integrated into a clinical pathway for FND in time. It is also being considered for modification so it can be used as the basis for a transdiagnostic intervention for other functional disorders such as IBS and fibromyalgia.

The study resulted in resources being developed; Two of the neurologists contributed by agreeing to be filmed talking about FND. This has resulted in patient information media for those receiving a diagnosis which can be accessed on NHS Grampian's web pages. The intervention was also very well received by those taking

part with benefits in health beliefs and quality of life emerging from only two sessions, although care has to be taken in attributing these differences to the intervention as there is no comparison group.

The high attrition rate and small sample size are obvious limitations. However numbers in both these areas are in line with similar research. In research it is common for an intervention to have a team of people working on it. In this study there was one person [the researcher (PI)] responsible for coordinating all recruitment contacts, administration, completing all the baseline appointments, inputting and analysing the data. There were different lengths of time that people may have had to wait before the group started. Potentially symptoms may have improved and the person decided not to attend, alternatively they may have deteriorated and people felt too unwell. As this was a research project it was not possible to contact patients to ask their reasons for non-attendance. Future research would add to the data collected in the feasibility project which may augment the benefits seen in participants to date. Some feedback highlighted that there was some difficulty in participants' ability to remember the information later due to cognitive difficulties. Developing patient resources based on the group content and format would address this limitation of the study.

Future research

The current study did not have a comparison group, randomised control trials (RCT) are considered the 'gold standard' when evaluating interventions. The efficacy of a brief intervention for FND may benefit from the RCT design, however matching the heterogeneity of participants within the FND population may be challenging. Future research would also benefit from running the intervention on a larger scale in NHS Grampian with a larger study team. More staff would increase capacity for

completing assessment and allow groups to be run sooner after baseline. A larger study to explore if the changes seen within the outcome measures in the feasibility study are retained would be beneficial. This would allow opportunity to collect further data on health beliefs and quality of life to explore if there is a predictive utility regarding who is likely to attend based on the personal control and physical health limitation questions. Running a larger study would also allow more data for the EQ-5D and allow QALY adjusted life years to be calculated to inform cost effectiveness of the group intervention.

Significant differences between attenders and non-attenders were detected in the domains of health beliefs and quality of life, suggesting that those not attending were less confident in their ability to manage their condition, feeling more physically restricted. There may also have been a belief that the intervention may not be suitable. No significant differences between access to healthcare sectors summary costs, however there was evidence of high utilisation across sectors with primary care being accessed the most suggesting that supporting GPS to help people manage the condition may be worth exploring.

For those completing the intervention symptom levels for cognition mood remained stable. Significant improvements were seen in levels of fatigue/energy on the SF-36 at follow up. Greater illness understanding was reported post group that which remained at follow up. Suggesting a period of adjustment is beneficial to allow participants to put knowledge into action.

Although there were no significant differences on healthcare utilisation measures, 63% of completers showed improvements in their health states between T1 and T4 as indexed by the EQ-5D-5L. This showed encouraging results from this

ambitious feasibility project. Participants welcomed the intervention, feeling it exceeded their expectations and resulted in significant improvements in their knowledge of their condition.

Conclusions

Results suggest this intervention is a feasible project to introduce further exploration of a post diagnosis intervention for those receiving a diagnosis of FND within FND Grampian. Sample sizes were small, as a result the magnitude of the effect size in the analyses was unable to be calculated on the current sample. Overall even with a small sample size and reduced power some promising results emerged that showed differences in the subscales of health beliefs and quality of life measures for attenders and non-attenders as well as completers. A larger study could explore if these subscales hold validity in predicting who is likely to attend interventions as well as continuing to assess if the content met the needs of the participants.

References

- Barrett-Naylor R., Gresswell D.M., & Dawson, D. L. (2018). The effectiveness and acceptability of a guided self-help acceptance and commitment therapy (ACT) intervention for psychogenic nonepileptic seizures. *Epilepsy and Behavior*, 88, 332-340.
- Barry, J., Wittenberg, D., Bullock, K., Michaels, J., Classen, C., & Fisher, R. (2008). Group therapy for patients with psychogenic nonepileptic seizures: A pilot study. *Epilepsy & Behavior*, 13(4), 624-629.
- Baslet, G., Dworetzky, B., Perez, D. L., & Oser, M. (2015). Treatment of psychogenic nonepileptic seizures: Updated review and findings from a mindfulness-based intervention case series. *Clinical EEG and Neuroscience*, 46(1), 54-64.
- Blake, J., Ablitt, A., Ruffmann, C., Morley, K., William, S., Moore, A., et al. (2019). Functional neurological disorder: Reflections on a pilot psychoeducational group in Hampshire. *The Neuropsychologist*, 8, 28-33.
- BPS Division of Neuropsychology. (2013). Management of patients with neurological medically unexplained symptoms within clinical services in NHS Scotland: The role of neuropsychology , *BPS, Scotland*,
- Broadbent, E., Petrie, K. J., Main, J., & Weinman, J. (2006). The brief illness perception questionnaire. *Journal of Psychosomatic Research*, 60(6), 631-637.

- Carson, A., & Lehn, A. (2016). Epidemiology. In M. Hallett, J. Stone & A. Carson (Eds.), *Handbook of clinical neurology* (pp. 147-160) Academic Press.
- Chisholm, D., Knapp, M. R. J., Knudsen, H. C., Amaddeo, F., Gaite, L., Van Wijngaarden, B., et al. (2000). Client socio-demographic and service receipt inventory–European version: Development of an instrument for international research: EPSILON study 5. *The British Journal of Psychiatry*, 177(S39), s28-s33.
- Clark, D. M. (2011). Implementing NICE guidelines for the psychological treatment of depression and anxiety disorders: the IAPT experience. *International review of psychiatry*, 23, 318-327.
- Cope, S. R., Smith, J. G., King, T., & Agrawal, N. (2017). Evaluation of a pilot innovative cognitive-behavioral therapy-based psychoeducation group treatment for functional non-epileptic attacks. *Epilepsy & Behavior*, 70, 238-244.
- de Rooij, A., de Boer, M. R., van der Leeden, M., Roorda, P. D., Steultjens, M. P., & Dekker, J. (2014). Cognitive mechanisms of change in multidisciplinary treatment of patients with chronic widespread pain: A prospective cohort study. *Journal of Rehabilitation Medicine*, 46(2), 173-180.
- Demartini, B., Batla, A., Petrochilos, P., Fisher, L., Edwards, M. J., & Joyce, E. (2014). Multidisciplinary treatment for functional neurological symptoms: A prospective study. *Journal of Neurology*, 261(12), 2370-2377.
- Devlin, N., Parkin, D., & Browne, J. (2009). Using the EQ-5D as a performance measurement tool in the NHS.

- Donkin, L., Ellis, C. J., Powell, R., Broadbent, E., Gamble, G., & Petrie, K. J. (2006). Illness perceptions predict reassurance following a negative exercise stress testing result. *Psychology and Health*, 21(4), 421-430.
- Edwards, M. J., & Bhatia, K. P. (2012). Functional (psychogenic) movement disorders: Merging mind and brain. *The Lancet Neurology*, 11(3), 250-260.
- Field, A. (2005). *Discovering statistics using IBM SPSS statistics* sage.
- Gelauff J.M., Carson A., Ludwig L., Tijssen M.A.J., & Stone, J. (2019). The prognosis of functional limb weakness: A 14-year case-control study. *Brain*, 142(7), 2137-2148.
- Glattacker, M., Opitz, U., & Jäckel, W. (2010). Illness representations in women with fibromyalgia. *British Journal of Health Psychology*, 15(2), 367-387.
- Goldstein, L. H., Deale, A. C., O'Malley, S. J. M., Toone, B. K., & Mellers, J. D. (2004). An evaluation of cognitive behavioral therapy as a treatment for dissociative seizures: A pilot study. *Cognitive and Behavioral Neurology*, 17(1), 41-49.
- Hays, R. D., & Morales, L. S. (2001). The RAND-36 measure of health-related quality of life. *Annals of Medicine*, 33(5), 350-357.
- Healthcare Improvement Scotland. (2012). Stepped care for functional neurological symptoms: A new approach to improving outcomes for a common neurological problem in Scotland. *Qis*,

- Herdman, M., Gudex, C., Lloyd, A., Janssen, M., Kind, P., Parkin, D., et al. (2011). Development and preliminary testing of the new five-level version of EQ-5D (EQ-5D-5L). *Quality of Life Research*, 20(10), 1727-1736.
- Jenkinson, C., Wright, L., & Coulter, A. (1994). Criterion validity and reliability of the SF-36 in a population sample. *Quality of Life Research*, 3(1), 7-12.
- Kendrick, T., & Pilling, S. (2012). Common mental health disorders--identification and pathways to care: NICE clinical guideline. *The British Journal of General Practice : The Journal of the Royal College of General Practitioners*, 62(594), 47-49.
- Knoop, H., Prins, J. B., Moss-Morris, R., & Bleijenberg, G. (2010). The central role of cognitive processes in the perpetuation of chronic fatigue syndrome. *Journal of Psychosomatic Research*, 68(5), 489-494.
- Kroenke, K., Spitzer, R. L., Williams, J. B., & Löwe, B. (2010). The patient health questionnaire somatic, anxiety, and depressive symptom scales: A systematic review. *General Hospital Psychiatry*, 32(4), 345-359.
- Leventhal, H., Forster, R., Leventhal, E., Aldwin, C., Park, C., & Spiro, A. (2007). Self-regulation of health threats, affect, and the self. *Handbook of Health Psychology and Aging*, , 341-366.
- Leventhal, H., Halm, E., Horowitz, C., Leventhal, E. A., Ozakinci, G., Sutton, S., et al. (2004). The SAGE handbook of health psychology.

- Leventhal, H., Meyer, D., & Nerenz, D. (1980). The commonsense representation of illness danger. *Contributions to Medical Psychology*, 2, 7-30.
- Leventhal, H., Nerenz, D., & Steele, D. (1984). Illness representations and coping with health threats in Baum A, Taylor SE, Singer JE, editors. *Handbook of psychology and health. volume IV: Social psychological aspects of health*.
- Libbon, R., Gadbaw, J., Watson, M., Rothberg, B., Sillau, S., Heru, A., et al. (2019). The feasibility of a multidisciplinary group therapy clinic for the treatment of nonepileptic seizures. *Epilepsy & Behavior*, 98, 117-123.
- Lutz, M., & Helmstaedter, C. (2005). EpiTrack: Tracking cognitive side effects of medication on attention and executive functions in patients with epilepsy. *Epilepsy & Behavior*, 7(4), 708-714.
- Monzoni, C. M., Duncan, R., Grünewald, R., & Reuber, M. (2011). How do neurologists discuss functional symptoms with their patients: A conversation analytic study. *Journal of Psychosomatic Research*, 71(6), 377-383.
- National Institute for Health and Care Excellence. (2019). Suspected neurological conditions: Recognition and referral.
- Nettleton, S., O'Malley, L., Watt, I., & Duffey, P. (2004). Enigmatic illness: Narratives of patients who live with medically unexplained symptoms. *Social Theory & Health*, 2(1), 47-66.

- Nielsen, G., Ricciardi, L., Demartini, B., Hunter, R., Joyce, E., & Edwards, M. (2015). Outcomes of a 5-day physiotherapy programme for functional (psychogenic) motor disorders. *Journal of Neurology*, 262(3), 674-681.
- Osborne, J. W., & Overbay, A. (2004). The power of outliers (and why researchers should always check for them). *Practical Assessment, Research, and Evaluation*, 9(1), 6.
- Parees, I., Kassavetis, P., Saifee, T. A., Sadnicka, A., Bhatia, K. P., Fotopoulou, A., et al. (2012). 'Jumping to conclusions' bias in functional movement disorders. *Journal of Neurology, Neurosurgery, and Psychiatry*, 83(4), 460-463.
- Pennington, C., Newson, M., Hayre, A., & Coulthard, E. (2015). Functional cognitive disorder: What is it and what to do about it? *Practical Neurology*, 15(6), 436-444.
- Pick, S., Anderson, D. G., Asadi-Pooya, A. A., Aybek, S., Baslet, G., Bloem, B. R., et al. (2020). Outcome measurement in functional neurological disorder: A systematic review and recommendations. *Journal of Neurology, Neurosurgery, and Psychiatry*,
- Prigatano, G. P., Stonnington, C. M., & Fisher, R. S. (2002). Psychological factors in the genesis and management of nonepileptic seizures: Clinical observations. *Epilepsy & Behavior*, 3(4), 343-349.
- Rommelfanger, K. S., Factor, S. A., LaRoche, S., Rosen, P., Young, R., & Rapaport, M. H. (2017). Disentangling stigma from functional neurological disorders: Conference report and roadmap for the future. *Frontiers in Neurology*, 8, 106.

- Rusch, M. D., Morris, G. L., Allen, L., & Lathrop, L. (2001). Psychological treatment of nonepileptic events. *Epilepsy & Behavior*, 2(3), 277-283.
- Rutter, C. L., & Rutter, D. R. (2002). Illness representation, coping and outcome in irritable bowel syndrome (IBS). *British Journal of Health Psychology*, 7(4), 377-391.
- Sassi, F. (2006). Calculating QALYs, comparing QALY and DALY calculations. *Health Policy and Planning*, 21(5), 402-408.
- Sharpe, M., Stone, J., Hibberd, C., Warlow, C., Duncan, R., Coleman, R., et al. (2010). Neurology out-patients with symptoms unexplained by disease: Illness beliefs and financial benefits predict 1-year outcome. *Psychological Medicine*, 40(4), 689-698.
- Sharpe, M., Walker, J., Williams, C., Stone, J., Cavanagh, J., Murray, G., et al. (2011). Guided self-help for functional (psychogenic) symptoms: A randomized controlled efficacy trial. *Neurology*, 77(6), 564-572.
- Siegrist, J., & Junge, A. (1989). Conceptual and methodological problems in research on the quality of life in clinical medicine. *Social Science & Medicine*, 29(3), 463-468.
- Spitzer, R. L., Kroenke, K., Williams, J. B., & Löwe, B. (2006). A brief measure for assessing generalized anxiety disorder: The GAD-7. *Archives of Internal Medicine*, 166(10), 1092-1097.

- Stone, J., Carson, A., & Sharpe, M. (2005). Functional symptoms and signs in neurology: Assessment and diagnosis. *Journal of Neurology, Neurosurgery, and Psychiatry*, 76 Suppl 1, i2-12.
- Teodoro, T., Edwards, M. J., & Isaacs, J. D. (2018). A unifying theory for cognitive abnormalities in functional neurological disorders, fibromyalgia and chronic fatigue syndrome: Systematic review. *Journal of Neurology, Neurosurgery, and Psychiatry*, 89(12), 1308-1319.
- Torrance, G. W. (1987). Utility approach to measuring health-related quality of life. *Journal of Chronic Diseases*, 40(6), 593-600.
- van Wilgen, C. P., van Ittersum, M. W., Kaptein, A. A., & van Wijhe, M. (2008). Illness perceptions in patients with fibromyalgia and their relationship to quality of life and catastrophizing. *Arthritis & Rheumatism: Official Journal of the American College of Rheumatology*, 58(11), 3618-3626.
- Věchetová, G., Slovák, M., Kemlink, D., Hanzlíková, Z., Dušek, P., Nikolai, T., et al. (2018). The impact of non-motor symptoms on the health-related quality of life in patients with functional movement disorders. *Journal of Psychosomatic Research*, 115, 32-37.
- Ware Jr, J. E., & Sherbourne, C. D. (1992). The MOS 36-item short-form health survey (SF-36): I. conceptual framework and item selection. *Medical Care*, 473-483.

- Yang, F., Devlin, N., & Luo, N. (2019). Impact of mapped EQ-5D utilities on cost-effectiveness analysis: In the case of dialysis treatments. *The European Journal of Health Economics*, 20(1), 99-105.
- Zaroff, C. M., Myers, L., Barr, W. B., Luciano, D., & Devinsky, O. (2004). Group psychoeducation as treatment for psychological nonepileptic seizures. *Epilepsy & Behavior*, 5(4), 587-592.

Chapter 2: Appendices

Appendix 1:Correlational Table for symptom duration and psychological variables

Pearson correlations between duration of symptoms and scores on measures of Cognitive Function, Mood Quality of life, and Health beliefs at baseline

Psychological Domain	Baseline Outcome Measures	All	Attendees	Non-Attendees
		r (n=29)	r (n=16)	r (n=13)
Cognitive Function	Epitrak Score	.31	.33	-.25
Mood	PHQ9 Depression	.04	.07	-.03
	GAD7 Anxiety	-.29	-.33	-.25
HRQoL	EQ-5D VAS	-.04	-.19	.38
	SF36 Physical Function	-.26	-.37	-.08
	SF36 Physical health limitations	-.16	-.23	.39
	SF36 Emotional health limitations	.27	.35	.13
	SF36 Energy/Fatigue	-.07	-.29	.47
	SF36 Emotional Wellbeing	.36	.39	.30
	SF36 Social Function	.01	.04	-.07
	SF36 Pain	-.06	-.24	.19
	SF36 General Health	.01	-.20	.40
Health Beliefs	IPQ-B Consequences	.04	.11	-.16
	IPQ-B Timeline	.39*	.34	.51
	IPQ-B Personal Control	.09	.12	.10
	IPQ-B Treatment Control	.15	.31	-.18
	IPQ-B Identity	.10	.15	-.07
	IPQ-B Illness Concern	.04	-.01	.16
	IPQ-B Coherence	.32	.40	.08
	IPQ-B Emotional Representation	-.35	-.35	-.36
	IPQ-B Level of threat	-.16	-.18	-.14



THE UNIVERSITY
of EDINBURGH

Consent Form

Study: A Feasibility Study for a Psychoeducation Group for individuals with a Functional Neurological Disorder

Please initial

1. I confirm that I have read and understand the information sheet Version No: 2 Date: 07/01/19 for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my medical care or legal rights being affected. Data collected up until the point of withdrawal may still be used in analysis.
3. I understand that relevant sections of my medical notes and data collected during the study may be looked at by individuals from the sponsor (University of Edinburgh), from regulatory authorities if appropriate, or from the NHS Board/Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.
4. I agree to my GP being informed of my participation in the study.
5. I understand that anonymised quotations from my feedback may be used for presentations and publications.
6. I agree to be sent a summary of the research results when the study is complete
7. I agree to be contacted by the study team for future studies that they may be undertaking. I understand identifiable contact information will be kept after the end of this study and this information will be held in accordance with the data protection act.
8. I agree to take part in the above study.

Name of participant

Date

Signature

Name of person taking consent

Date

Signature

Original x 1 to be retained in site file. Copy x 1 to participant



THE UNIVERSITY
of EDINBURGH

Participant Information Sheet

Study: A Feasibility Study for a Psychoeducation Group for
Individuals with a Functional Neurological Disorder

Introduction

This sheet is about a research study that is happening in NHS Grampian. It gives some information to help you decide if you want to take part or not. Please read this sheet and speak about it with others if you wish. If you have questions, please get in touch with Dr Pauline Insch or Dr Fiona Summers on 01224 559352. We are happy to answer any questions and would like to thank you for taking time to read this information.

What is this study about?

At the moment we do not have any psychological treatments that have been designed to meet the needs of people who have been diagnosed with a Functional Neurological Disorder. In this research we are investigating how useful it is to give patients some additional information to help them to explain and manage their symptoms. This will take the form of a small group which will have two sessions, these will be around two weeks apart and each session will last for approximately 90 minutes. At present these group sessions are only available to participants taking part in the research study.

Why have I been asked to take part?

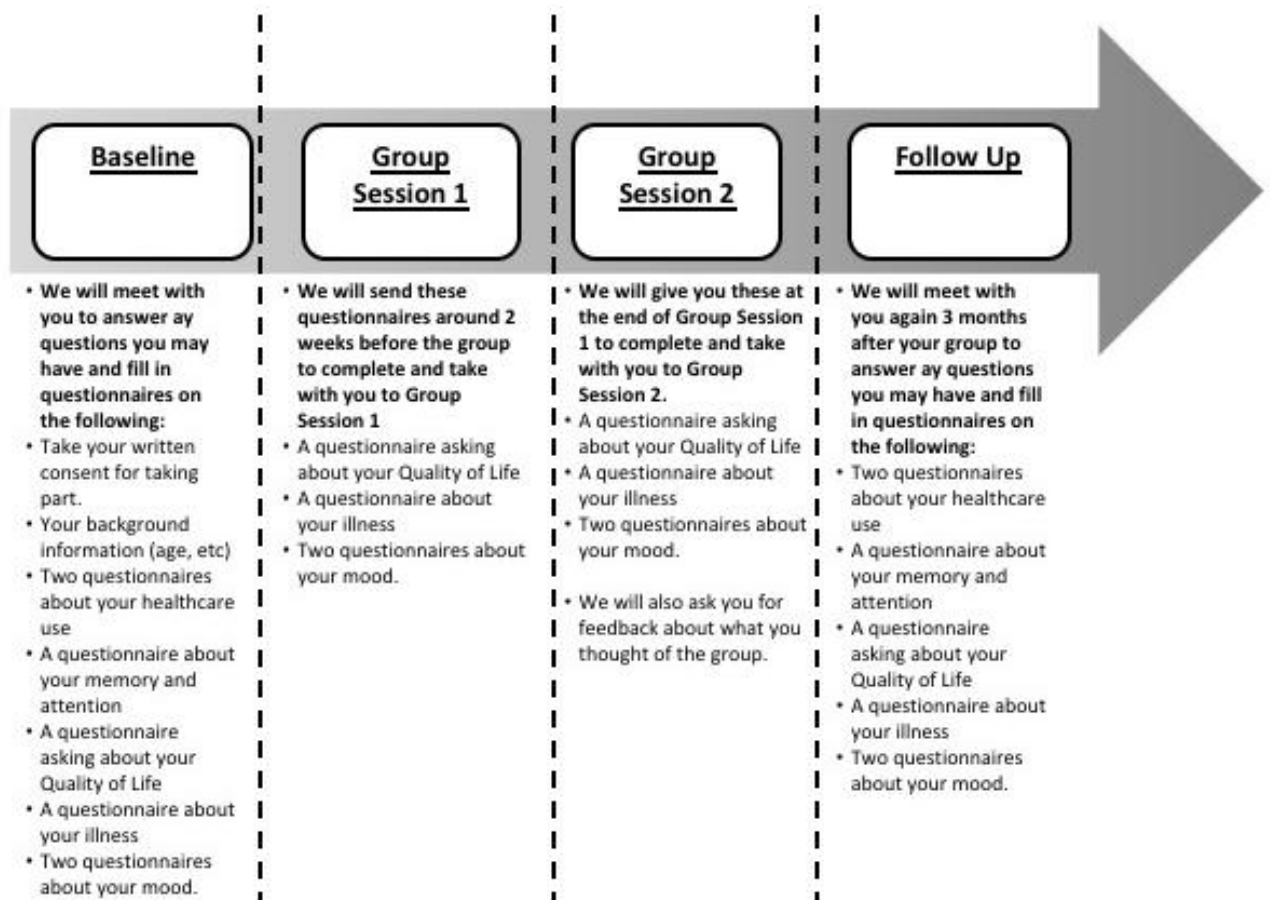
You have been asked as you have been diagnosed with a Functional Neurological Disorder by a neurologist in the Department of Neurology, Aberdeen Royal Infirmary.

Do I have to take part?

No. It is up to you to make up your mind if you want to take part or not. If you want to take part, please return the study reply sheet and Dr Pauline Insch will contact you. If you decide to take part, and then change your mind you can come out of the study at any time and do not have to tell us why. If you decide you do not want to take part, or take part then change your mind later, this will not change the care you receive.

What will happen to me if I take part?

The flowchart below shows what taking part in this study would involve. Please see appendix A for more information on the questionnaires and approximate time these will take.



Once we receive your reply form Dr Pauline Insch will contact you to arrange an appointment that is convenient for you. This appointment will take around 60 minutes, you will have the chance to ask any questions you may have and if you are happy to go ahead, you will be asked to sign a consent form to give us permission to use your information in the research and complete some questionnaires. Two weeks before the group we will send you questionnaires to complete and take with you to the group session. The questionnaires are short and take around 20-30 minutes on average in total. There will be two groups sessions, each will last around 90 minutes and be two weeks apart. At the last session you will be asked to complete questionnaires and to give us your feedback on the group you have attended. Three months after the group has ended, we will contact you to arrange a final appointment to answer any questions you may have and complete the final set of questionnaires.

What will the group be like?

If you decide to take part you will be asked to attend 2 group sessions spaced a fortnight apart which last for approximately 90 minutes. There will be around 6 people in each group.

There is no requirement for you to speak about or share your experiences in the group or with others attending. The purpose of the group is to provide information that will hopefully allow you to understand and manage difficulties associated with your illness. Information on what the group sessions will cover is in the table below

<u>Session</u>	<u>Key Topics</u>
1 Introduction to FND	<ul style="list-style-type: none">▪ What is FND? Making sense of symptoms▪ The Brain-Body-Thought Link
2 Moving Forward with FND	<ul style="list-style-type: none">▪ Moving forward: Managing symptoms▪ Obstacles to recovery▪ Signposting to helpful resources

The group aims to help you to:

- Understand more about what is meant by functional neurological disorder
- Develop knowledge about how physical and psychological processes interact in the body
- Build knowledge of effective coping strategies and things that may be less helpful.
- Gain awareness of resources available
- Have opportunity to meet with others who have been through similar experiences.

At the end of the second session we will ask you for your feedback about the group this feedback will be questionnaire based and anonymous .

What are the possible disadvantages and risks if I take part?

It is not likely any harm will come to you if you take part. If however, you feel concerned or upset about any issue raised in the questionnaires or in the group content, there will be a consultant clinical neuropsychologist present, Dr Fiona Summers, within the group sessions to assist you if this is necessary. Information on managing emotions will also form part of the group programme. You can also talk to Dr Pauline Inch or either in the appointment, after the group or on 01224 559352. The project team will be happy to discuss any concerns you may have.

It may be an inconvenience on your time to complete the questionnaires used in the research (see Appendix A at the end of this information sheet, for more information); however, we have made every effort to keep these to a minimum and will make every effort to schedule the group and the one to one sessions at times convenient to you.

What are the possible benefits if I take part?

We cannot promise that you will find the group helpful, but we hope having more information about your diagnosis and meeting others with Functional Neurological Disorders may be beneficial to you and help you manage your symptoms. Although completing the questionnaires outlined in Appendix A may be an additional time commitment for you, we hope the information collected from these will help us understand more about some of the things patients face when we are developing our treatment options for those that have a Functional Neurological Disorder as there is very little information available to clinicians at present.

Will my taking part in this study be kept confidential?

All information which is collected about you during the course of the research will be kept strictly confidential. This information will be coded so no personal details will be available from the questionnaires alone, all personal information is kept separate from study results. Procedures for storing data are compliant with the Data Protection Act (2018). The identifiable information shall be stored separately from the questionnaires also in a locked filing cabinet in NHS Grampian. Electronic data will only be stored on NHS computer server, computers are password protected as per local NHS board policy. Only the research team will be able to identify participants during the course of the research.

Will my you contact my General Practitioner (GP)?

We will ask your permission to inform your GP that you are taking part in this feasibility study. If you give permission, we will send your GP a copy of this an information sheet giving information on this feasibility study and what taking part involves for you. You can find out more about how we use your information and our legal basis for doing so in our Privacy Notice at: <https://www.ed.ac.uk/records-management/privacy-notice-research>

What happens to the results of the research study?

The results of this study will be submitted as doctoral thesis as part of Dr Pauline Insch's Doctorate in Clinical Psychology training. In addition, we hope the results will improve the service we offer patients within the Neurology Department. We may also publish the results in scientific journals and also present the results at conferences. However, no one will be able to recognise you in any report/publication or presentation.

Who is organising and funding the research?

The project is part of Dr Pauline Insch's training as part of the Doctorate in Clinical Psychology at the University of Edinburgh. The research has been sponsored by The University of Edinburgh and organised by the Department of Clinical Neuropsychology at Aberdeen Royal Infirmary. No additional funding has been provided.

Who reviewed the study?

The North East of Scotland Research Ethics Service REC (2), which has responsibility for scrutinising NHS research proposals, has reviewed this study and raised no objections from the point of view of research ethics. It is a requirement that all relevant research records are made available for scrutiny by monitors from NHS Grampian, whose role is to check that the research is properly conducted and the interests of those taking part are protected.

Study Results and Future Research

We would like to send you information about the results of the study when the research is complete. We would also like your permission to send information about further research that may be of interest to you. These permissions are in the consent form, there is no obligation to receive information or hear about potential future research you can still participate if you don't wish to be kept informed or take part in other studies.

Contact for further information

If you have any questions you can contact Dr Fiona Summers or Dr Pauline Insch on 01224 559352.

If you would like to speak to someone independent of the study, please contact Dr Jackie Hamilton on 01224 559352.

You can also email the study team at nhsg.neuropsych@nhs.net

If you wish to make a complaint about the study you can do this through the usual NHS process by contacting the NHS Grampian Feedback service Summerfield House, 2 Eday Road, Aberdeen, AB15 6RE, by telephone (0345 337 6338) or by e-mail (nhsgrampian.feedback@nhs.net)

If you wish to raise a complaint on how we have handled your personal data, you can contact our Data Protection Officer who will investigate the matter. If you are not satisfied with our response or believe we are processing your personal data in a way that is not lawful you can complain to the Information Commissioner's Office (ICO) at <https://ico.org.uk/>.

Data Protection Officer contact information: University of Edinburgh, Data Protection Officer, Governance and Strategic Planning, University of Edinburgh, Old College, Edinburgh, EH8 9YL, by telephone 0131 651 4114 or by email dpo@ed.ac.uk

Thank you for taking time to read about our project

- If you want to take part, please complete the accompanying reply form and return in the pre-paid envelope provided
- If you do not wish to participate then please choose the option on the enclosed reply form and return in the pre-paid envelope provided.
- If you do not wish to participate or return the forms, then dispose of as you see fit.

Information sheet Appendix A Study Questionnaires and approximate completion times

Baseline Session (One to One)		
Questionnaire	Description	Time to complete (approximately)
Informed Consent	A form to get permission from you to take part in the research	5mins or less
Demographics	A form to get background information e.g. your age and gender	5mins or less
EQ5D	A questionnaire about your quality of life related to your health	5mins or less
Client Services Receipt Inventory (CSRI)	A questionnaire about the Health services you use related to your illness	15 mins or less
Epi Trak	A questionnaire about memory and attention	10 mins or less
Short Form 36 (SF36)	A questionnaire about your quality of life related to your health	10 mins or less
Brief Illness Perception Questionnaire (B-IPQ)	A questionnaire about what you think about your illness	5mins or less
Generalised Anxiety Disorder 7 (GAD7)	A questionnaire about Anxiety	5mins or less
Patient Health Questionnaire (PHQ9)	A questionnaire about Depression	5mins or less
Approximate time for Baseline Questionnaires		65 mins or less
Pre-Group Session 1		
Short Form 36 (SF36)	A questionnaire about your quality of life related to your health	10 mins or less
Brief Illness Perception Questionnaire (B-IPQ)	A questionnaire about what you think about your illness	5mins or less
Generalised Anxiety Disorder 7 (GAD7)	A questionnaire about Anxiety	5mins or less
Patient Health Questionnaire (PHQ9)	A questionnaire about Depression	5mins or less
Approximate time for Pre-Group Session 1 Questionnaires		25 mins or less

Post Group Session 2		
Questionnaire	Description	Time to complete (approx.)
Short Form 36 (SF36)	A questionnaire about your quality of life related to your health	10 mins or less
Brief Illness Perception Questionnaire (B-IPQ)	A questionnaire about what you think about your illness	5mins or less
Generalised Anxiety Disorder 7 (GAD7)	A questionnaire about Anxiety	5mins or less
Patient Health Questionnaire (PHQ9)	A questionnaire about Depression	5mins or less
Group Feedback	A form asking for your thoughts on the group programme	5mins or less
Approximate time for Post Group Session 2 Questionnaires		30 mins or less
Follow Up Session 3 months after group (One to One)		
EQ5D	A questionnaire about your quality of life related to your health	5mins or less
Client Services Receipt Inventory (CSRI)	A questionnaire about the Health services you use, and other costs related to your illness	15 mins or less
Epi Trak	A questionnaire about memory and attention	10 mins or less
Short Form 36 (SF36)	A questionnaire about your quality of life related to your health	10 mins or less
Brief Illness Perception Questionnaire (B-IPQ)	A questionnaire about what you think about your illness	5mins or less
Generalised Anxiety Disorder 7 (GAD7)	A questionnaire about Anxiety	5mins or less
Patient Health Questionnaire (PHQ9)	A questionnaire about Depression	5mins or less
Approximate time for follow up session questionnaires		55 mins or less

Department of Clinical Neuropsychology
Pre-Group Evaluation



What are your expectations of attending the FND group?

Please mark the box that matches your level of knowledge before the group:

	I know a lot	I know a fair bit	I know some things	I don't know much	I know nothing
What FND is.					
How FND is diagnosed					
How Physical and Psychological processes interact					
What effective coping strategies are					
What can make symptoms worse					
What resources are available					

	Very Helpful	Helpful	Neutral	Unhelpful	Very Unhelpful
How helpful do you think it will be to meet with others who have been through similar experiences?					

Thank you for your Feedback.

Department of Clinical Neuropsychology
Post-Group Evaluation



ID Number:

Workshop Date:

Were your expectations of the group met?

Please mark the box that matches your level of knowledge now you have completed the group:

	I know a lot	I know a fair bit	I know some things	I don't know much	I know nothing
What FND is.					
How FND is diagnosed					
How Physical and Psychological processes interact					
What effective coping strategies are					
What can make symptoms worse					
What resources are available					

	Very Helpful	Helpful	Neutral	Unhelpful	Very Unhelpful
I found meeting others who have been through similar experiences.					

Thank you for your Feedback.

Appendix 5: Participant Feedback



THE UNIVERSITY
of EDINBURGH

Group Evaluation Form

Please complete the evaluation form before you leave the final session. Your views will help us review and improve the group for future participants. Please circle the answer you agree with most, there is space for comments if you wish to give more information.

1. The session lasted for 1.5 hours, was this...

Much too long	A little long	About right	A little short	Much too short
---------------	---------------	-------------	----------------	----------------

Comments:

2. There were 2 sessions in total, was this...

Too many	A little too many	About right	Not quite enough	Definitely not enough
----------	-------------------	-------------	------------------	-----------------------

Comments:

3. The sessions were spaced a fortnight apart, was this...

Too often	A little too often	About right	Not quite often enough	Definitely not often enough
-----------	--------------------	-------------	------------------------	-----------------------------

Comments:

If you chose too close or too far apart how much time apart do you feel would have been better?

4. The range of topics covered was

Too many	A little too many	About right	Not quite enough	Definitely not enough
----------	-------------------	-------------	------------------	-----------------------

Comments:

5. The amount of information covered in each session was

Too much	A little too much	About right	Not quite enough	Definitely not enough
----------	-------------------	-------------	------------------	-----------------------

Comments:

6. How comfortable was the room and the facilities where the group took place

Very comfortable	Quite comfortable	Adequate	Quite poor	Very poor
------------------	-------------------	----------	------------	-----------

Comments:

Please Turn Over

7. How approachable were the group facilitators?

Very easy to talk to	Quite easy to talk to	Adequate	Somewhat difficult to talk to	Very hard to talk to
----------------------	-----------------------	----------	-------------------------------	----------------------

Comments:

8. How easy was the information to understand and remember?

Very easy	Quite easy	Neither easy or difficult	Quite difficult	Very difficult
-----------	------------	---------------------------	-----------------	----------------

Comments:

9. Was there anything about the sessions that you particularly liked?

10. Was there anything about the sessions you would change?

11. Would you recommend this group programme to a friend?

☐ YES

☐ NO

If you wish to comment further please do so below.

Thank you for your feedback.

Appendix 6: Demographic Information



THE UNIVERSITY
of EDINBURGH

Dr Pauline Insch (Chief Investigator) Telephone: 01224 559352
Department of Clinical Neuropsychology
Ashgrove House
Aberdeen Royal Infirmary
Aberdeen
AB25 2NZ

Participant ID No _____

Date Completed _____

Study Stage Baseline

Demographics

Study: A Feasibility Study for a Psychoeducation Group for Functional Neurological Disorders

Meets Inclusion Criteria

☐ (tick to confirm) initials/Date ____/____-____-____

1. D.O.B

/ /

2. Age

3. Postcode

4. SIMD

5. Gender

Male ☐ Female ☐

6. Date invited

/ /

7. Date Reply Received

/ /

8. Date Contacted

/ /

9. Date of Baseline Appointment

/ /

10. Marital Status ☐ Married/Partner ☐ Single ☐ Widowed ☐ Divorced/Separated

11. Employment Status ☐ F/T Work ☐ P/T Work ☐ Retired ☐ Not Working (Health)
☐ Medically Retired ☐ Student

12. Education

Years at primary school Years at secondary school Age left school

Number of years in higher or further education

(Record current year of study if currently undertaking a degree)

13. Symptom Duration

Years Months

14. Age when symptoms began

Years

Appendix 7: Presenting Symptoms

Primary Presenting Symptom Categories

Sensory

Numbness, tingling in the face, body or limbs.
Pain in the face, body or limbs
Functional visual symptoms
Pain from Headache/Migraine

Cognitive

Memory Problems
Attention problems

Motor

Functional limb weakness/paralysis
Functional movement disorders; including tremor, spasms (dystonia), jerky movements (myoclonus) and problems walking (gait disorder)
Functional speech symptoms; including whispering speech (dysphonia), slurred or stuttering speech

Altered Awareness

Dissociative (non-epileptic) seizures
Blackouts
Collapse/faints

Non-Specific

Physical and psychological symptoms experienced by patients with FND
Fatigue
Sleep problems
Bowel and bladder problems
Anxiety and depression.
Eye problems

Appendix 8: Qualitative Summary of Participant Expectations from Pre & Post Group

Patient ID	Pre-Group Expectations	Post-Group Expectations
PT02	To help educate professionals about FND so that future FND patients know more about their disorder and not [feel] confused or feel fobbed off	Yes they were
PT03	To be very honest I have no expectations other than it will increase my awareness and perhaps help the FND cause in Grampian NHS	Yes. I found the group session to be very informative and reassuring. Well done for presenting professional yet friendly and relaxed sessions
PT08	Meet others and share experiences to do with FND. Get a better understanding on what FND actually is and possibly how to manage symptoms	I found the group really helpful for more of an understanding of FND
PT12	meet others like self and see how to cope on day and daily basis and see if anything can help	Yes - nice to see others with similar symptoms, gave understanding of why body is doing what [it is] doing and being given info' in finding others/body
PT13	To meet other people with the problems with FND	Yes. The group was very helpful, I realised I'm not alone
PT16	finding out what FND is and what is available to those who suffer from it. Maybe find link between FND and PTSD. What treatments (if any) are available	To be honest I did not have any expectations preferring to keep an open mind as I had went through similar groups relating to PTSD and was disappointed. I did however find this group very helpful, especially meeting others who suffer from FND
PT17	To understand more about my condition, learn how to manage my symptoms, learn what can make symptoms worse, find out what resources there are available, learn what lifestyle choices I can make	Yes it was great to be part of the study. The group sessions gave me peace of mind. I really appreciated running through coping strategies and the bit about what can make symptoms worse.
PT19	increase awareness to what it actually is. Listen to other people's understanding	They certainly were. I wasn't initially sure of what to expect but as it was a small group we got more input into it. Very informative.

PT23	How to better manage day to day life and have a better understanding of anatomy processes if available	Yes - well presented, good balance of info and personal discussion. Info presented to best of their knowledge. I understand its limited due to research
PT24	Mutual support and a sense of understanding. Opportunity to build in shared experiences to improve experiences for others coming to a diagnosis	Yes - made useful by good balance of info and experiences of others
PT25	I would like to find out more about how I feel and if there are others in the same position	My expectations of group are more than met - fantastic discussions about feelings and FND
PT26	How to move forward with FND, meeting peer group, shaping support for those with FND diagnosis in the future	Yes and more! The new FND leaflet is inspired.
PT28	Finding out more about FND. Meeting new people with the same diagnosis as yourself, meeting new people and making new friendships	I learned a lot about FND and what to do now I know it's not my fault and I'm not alone

Appendix 9: Qualitative Summary Participant Group Feedback

Q1 The session lasted for 1.5 hours was this...

Feedback Sheet No	Option	Comment
4	A little short	Feel as quite few slides maybe a Q+A at the end
6	A little short	Felt it couldn't lasted 2 hours maybe
7	A little short	Time was good in terms of illness, but felt there could be a lot more discussion about each other's experiences
8	A little short	Extend to two full ours with a 15 min break in between

Q2 There were 2 sessions in total was this...

1	Not quite enough	Group discussions were great, I would have liked more time to get to know them a little better.
4	Not quite enough	Think a third meeting would be good, of feel enough points to consider.
6	Not quite enough	I would prefer more, just as it was great for info. And others with similar.
8	About right	Alternatively have the two sessions in one day due to participants' long travel

Q3 The sessions were spaced a fortnight apart, was this...

2	Not quite often enough	Weekly sessions
4	Not quite often enough	Felt if it was weekly I would take it in better as my memory is bad.
11	Not quite often enough	A week apart

Q4 The range of topics covered was...

4	About right	Topics good, more time on each
---	-------------	--------------------------------

Q5 The amount covered in each session was..

2	About right	I would have preferred weekly sessions over 4-6 weeks to give a bit more time to take in and think about the discussions and recap and talk about them the following week.
---	-------------	--

Q6 How comfortable was the room and the facilities where the group took place ...

Feedback Sheet No	Option	Comment
2	Very Comfortable	Nice and warm friendly atmosphere
4	Very Comfortable	Room was fine

Q7 How approachable were the group facilitators..

2	Very easy to talk to	The information videos were very helpful and explained by the tutors present
4	Very easy to talk to	Very approachable
6	Very easy to talk to	Was very relaxed atmosphere and very helpful in discussions
7	Very easy to talk to	Very approachable facilitators

Q8 How easy was the information to understand and remember?

2	Quite easy	Tutors gave clear answers to questions
4	Very easy	Understand ok, remembering difficult.
6	Very easy	Very easy but my fault for not remembering
9	Neither easy nor difficult	Mainly because my memory isn't that great at the moment.

Q9 Was there anything about the group you particularly liked?

1	The friendliness, meeting the others and the professionals
2	The friendliness between the tutors and patients
3	The information and banter
4	The camaraderie and easily approachable psychologists
5	Everything
6	More people in the same boat as me and the helpful staff
7	Semi-structured and allowed for discussion
8	How informal and friendly it was. The facilitators would listen to your experiences
9	I like how at ease and interesting it was
10	Friendly open conversations. It was good to hear how others deal with FND.
11	I liked how FND was normalised and acknowledged that many GP's do not know about it

Q10 Was there anything about the sessions you would change?

- | | |
|----|---|
| 1 | A little more discussion amongst the group would be helpful but possibly not for everyone. |
| 2 | Spread over longer period maybe 4-6 weeks |
| 3 | No |
| 4 | No thought they were fine |
| 5 | No |
| 6 | Just more time for discussions really as can get in-depth |
| 7 | Add discussion times between participants whether in pairs or groups |
| 8 | No |
| 9 | Perhaps some more time for people to ask questions or tell experiences. Our group was quite small so might be harder in a larger group. |
| 10 | No |
| 11 | The videos were good but there was a lot of information to digest and interact with very quickly. |
-

Q11 Would you recommend this group to a friend

100% of participants said yes

Q12 Further comments

- | | |
|----|---|
| 1 | Great progress for patients and families dealing with FND
Felt very much at ease with tutors could ask them anything you were unsure of and had a lot of laughs with them. They were patient and friendly and answered everything that was asked of them. I found the sessions very helpful in many ways especially knowing I was not alone with my problems and I could do a lot to help myself and now I know how to deal with them. |
| 2 | |
| 3 | Overall pleased with the programme, it gave good insight into FND and gave further resources for once the programme completed. |
| 4 | Very interesting meeting people with the same diagnosis as yourself |
| 5 | It was great to have the chance of being part of such a complex study. It also helped me understand more about FND and the minefield that it comes with |
| 9 | I would definitely recommend to any others with FND or family etc |
| 10 | This was a well thought out group meeting where we learned a lot about FND that had not been explained to us when we were all diagnosed. I am really glad I had the opportunity to attend. |
-

Appendix 10: Ethical Approval NoSRES

North of Scotland Research Ethics Service

Summerfield House
2 Eday Road
Aberdeen
AB15 6RE

Telephone: 01224 558458
Facsimile: 01224 558609
Email: nosres@nhs.net



17 January 2019

Dr Pauline Inch
Trainee Clinical Psychologist
NHS Grampian
Department of Neuropsychology
Ashgrove House
Foresterhill Road
ABERDEEN
AB25 2ZN

Dear Dr Inch

Study title:	A feasibility study for a psychoeducation group intervention for people with Functional Neurological Disorder (FND)
REC reference:	18/NS/0137
Protocol number:	CAHSS1809/05
IRAS project ID:	247662

Thank you for your letter of 14 January 2019, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact hra.studyregistration@nhs.net outlining the reasons for your request.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise). Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System, at www.hra.nhs.uk or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	Version	Date
Covering letter on headed paper: Submission Covering Letter		15 November 2018
Covering letter on headed paper: Response to Provisional Opinion Letter		14 January 2019
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only)		24 July 2018
GP/consultant information sheets or letters: GP Letter	1	4 October 2018
IRAS Application Form	247662/1270551/37/99	16 November 2018
IRAS Checklist XML: Checklist 16012019		16 January 2019
Letters of invitation to participant: Reminder	1	13 September 2018
Letters of invitation to participant	2	7 January 2019
Non-validated questionnaire: Group Evaluation Form	1	13 September 2018
Non-validated questionnaire: Demographics	2	7 January 2019
Insurance Certificate		24 July 2018
Client Information Letter - Professional Indemnity Insurance		31 July 2018
Client Information Letter - Clinical Trial Liability Insurance		31 July 2018
Participant Consent Form	1	13 September 2018
Participant Information Sheet (PIS)	2	7 January 2019
Referee's report or other scientific critique report: Protocol Proposal Assessed by academics at university of Edinburgh		1 June 2018
Research protocol or project proposal	1	13 September 2018
Summary CV for Chief Investigator (CI): Pauline Inch		1 August 2018
Summary CV for Student: Pauline Inch		1 August 2018
Summary CV for Supervisor (student research): Clara Calia		21 July 2018
Validated questionnaire: EQ-5D-5L		16 November 2018*
Validated questionnaire: PHQ 9 and GAD 7		16 November 2018*
Validated questionnaire: SF36		16 November 2018*
Validated questionnaire: Brief - IPQ		16 November 2018*
Validated questionnaire: EpiTrack Test Protocol		February 2015
Validated questionnaire: Client Services Receipt Inventory	1	13 September 2018

* date received

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "*After ethical review – guidance for researchers*" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: <http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at <http://www.hra.nhs.uk/hra-training/>

18/NS/0137	Please quote this number on all correspondence
-------------------	---

With the Committee's best wishes for the success of this project.

Yours sincerely

Professor Helen Galley
Chair

Appendix 11: NHS Grampian Research and Development Approval

Research and Development

Foresterhill House Annexe
Foresterhill
ABERDEEN
AB25 2ZB



Dr Pauline Insch
NHS Grampian
Trainee Clinical Psychologist
Dept of Neuropsychology
Aberdeen Royal Infirmary
Aberdeen
AB25 2ZN

Date	18/01/2019
Project No	2018PC011
Enquiries to	Lynn / Louise
Extension	53846
Direct Line	01224 553846
Email	grampian.randdpermissions@nhs.net

Dear Dr Insch

Management Permission for Non-Commercial Research

STUDY TITLE:	A feasibility study for a psychoeducation group intervention for people with Functional Neurological Disorder (FND)
PROTOCOL NO:	V1; 13.9.18
REC REF:	18/NS/0137
R&D REF:	2018PC011

Thank you very much for sending all relevant documentation. I am pleased to confirm that the project is now registered with the NHS Grampian Research & Development Office. The project now has R & D Management Permission to proceed locally. This is based on the documents received from yourself and the relevant Approvals being in place.

All research with an NHS element is subject to the UK Policy Framework for Health and Social Care Research (2017 v3), and as Chief or Principal Investigator you should be fully committed to your responsibilities associated with this.

R&D Permission is granted on condition that:

- 1) The R&D Office will be notified and any relevant documents forwarded to us if any of the following occur:
 - Any Serious Breaches in Grampian (Please forward to pharmaco@abdn.ac.uk).
 - A change of Principal Investigator in Grampian or Chief Investigator.
 - Any change to funding or any additional funding
- 2) When the study ends, the R&D Office will be notified of the study end-date.
- 3) The Sponsor will notify all amendments to the relevant National Co-ordinating centre. For single centre studies, amendments should be notified to the R&D office directly.

We hope the project goes well, and if you need any help or advice relating to your R&D Management Permission, please do not hesitate to contact the office.

Yours sincerely

Susan Ridge
Non-Commercial Manager

cc: Sponsor – Charlotte Smith, Edinburgh
Research Monitor

Sponsor: University of Edinburgh

Appendix 12: Submission Guidelines for Neuropsychology



Other Electronic Access Options

Subscribe to PsycARTICLES on APA PsycNET® and access APA journals online

Pricing and Access

Editor: Keith Owen Yeates

ISSN: 0894-4105

eISSN: 1931-1559

Published: eight times, beginning in January

Impact Factor: 2.477

Psychology - Clinical: 44 of 130

5-Year Impact Factor: 3.200

Submission

Neuropsychology[®] is now using a software system to screen submitted content for similarity with other published content. The system compares each submitted manuscript against a database of 25+ million scholarly publications, as well as content appearing on the open web. This allows APA to check submissions for potential overlap with material previously published in scholarly journals (e.g., lifted or republished material). A similarity report will be generated by the system and provided to the *Neuropsychology* Editorial office for review immediately upon submission.

Starting in 2012, the completion of the [Author\(s\) Agreement Checklist \(PDF, 40KB\)](#) that signifies that authors have read this material and agree to adhere to the guidelines is now required. For new submissions, please be sure to include the submission checklist on the first page of your manuscript. Revisions do not need the checklist.

To submit to the Editorial Office of Keith Owen Yeates, please submit manuscripts electronically through the Manuscript Submission Portal in Microsoft Word or Open Office format.

The file must exactly copy, in all respects and in a single file, the complete APA-style printed version of the manuscript.

Authors with questions concerning manuscript submission should address these directly to the *Neuropsychology* Editorial Office.

In addition to addresses and phone numbers, please supply email addresses and fax numbers, if available, for potential use by the Editorial Office and later by the Production Office.

Keep a copy of the manuscript to guard against loss.

Neuropsychology is a peer-reviewed journal that typically publishes original research as full-length regular articles; systematic reviews and meta-analyses, as well as theoretical articles, are also welcome if they advance knowledge regarding human brain-behavior relationships. A detailed description of the editorial coverage policy appears on the inside of the front cover of each issue.

Other article formats, such as brief reports and case studies, will also be considered for publication.

Language

The official language of APA journals is English.

Neuropsychology frequently publishes manuscripts submitted by authors from non-English-speaking countries. Authors not fluent in English are strongly recommended to have their manuscript edited for English usage prior to submission. If this is not possible, a notation to this effect should be included in the cover letter to the editor.

Although time constraints prevent the editor and associate editors from assisting authors with their written English, several organizations have extended offers to the journal to provide this service for authors; contact the editor for more information.

Abstract and Keywords

Starting in 2010, all manuscripts published in *Neuropsychology* will include a structured abstract of up to 250 words. The Abstract, presented in paragraph form, should be typed on a separate page (page 2 of the manuscript), and must include each of the following sections:

Objective: A brief statement of the purpose of the study

Method: A detailed summary of the participants as well as descriptions of the study design, measures, and procedures

Results: A detailed summary of the primary findings that include effect sizes or confidence intervals with significance testing

Conclusions: A summary of the research and implications of the findings

After the abstract, please supply three to five keywords.

Key Points

Neuropsychology requires authors of all manuscripts to submit a short Key Points summary, written in conversational English, that summarizes the main takeaways for the article. The Key Points section allows authors' work to be more discoverable and easily interpreted by a number of audiences (clinicians, policy makers, news media).

In the manuscript, include a separate section called 'Key Points' after the Abstract.

Please structure the Key Points as follows; each section should be no more than one short sentence in length:

Question: What is the key question this paper addresses?

Findings: What are the primary findings?

Importance: What are the key scientific and practical implications of the findings?

Next Steps: What directions should be explored in future research?

Please refer to the [Guidance for Translational Abstracts and Public Significance Statements](#) page to help you write this text.

Manuscript Preparation

Until May 31st 2020, prepare manuscripts according to the [Publication Manual of the American Psychological Association](#) using the 6th or 7th edition. Starting June 1st 2020, all manuscripts should be submitted in the 7th edition. Manuscripts may be copyedited for bias-free language (see Chapter 3 of the 6th edition or Chapter 5 of the 7th edition).

Formatting instructions (all copy must be double-spaced) and instructions on preparing tables, figures, references, metrics, and abstracts appear in the *Manual*.

Also, all manuscripts are copyedited for bias-free language (see Chapter 3 of the *Manual*).

Visit the [Preparation and Submission](#) page in the Journals Publishing Resource Center for more information

Below are additional instructions regarding the preparation of display equations, computer code, and tables.

Tables

Use Word's Insert Table function when you create tables. Using spaces or tabs in your table will create problems when the table is typeset and may result in errors.

Review APA's [Journal Manuscript Preparation Guidelines](#) before submitting your article.

References

List references in alphabetical order. Each listed reference should be cited in text, and each text citation should be listed in the References section.

Examples of basic reference formats:

Journal Article:

Hughes, G., Desantis, A., & Waszak, F. (2013). Mechanisms of intentional binding and sensory attenuation: The role of temporal prediction, temporal control, identity prediction, and motor prediction. *Psychological Bulletin*, 139, 133–151.

<http://dx.doi.org/10.1037/a0028566>

Authored Book:

Rogers, T. T., & McClelland, J. L. (2004). *Semantic cognition: A parallel distributed processing approach*. Cambridge, MA: MIT Press.

Chapter in an Edited Book:

Gill, M. J., & Sypher, B. D. (2009). Workplace incivility and organizational trust. In P. Lutgen-Sandvik & B. D. Sypher (Eds.), *Destructive organizational communication: Processes, consequences, and constructive ways of organizing* (pp. 53–73). New York, NY: Taylor & Francis.